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Siblings of children and adults with intellectual and developmental disabilities:
psychological outcomes and sibling relationships

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List of Abbreviations

ID	Intellectual Disability
DD	Developmental Disability
IDD	Intellectual and/or Developmental Disability
SEND	Special Educational Need and/or Disability
SES	Socio-Economic Status
SEP	Socio-Economic Position
WLS	Wisconsin Longitudinal Study
MCS	Millennium Cohort Study

This thesis is dedicated to my brother.

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Declarations

This thesis has not been submitted for an award or degree at any university or institution. Where material has been derived from other sources, full bibliographical information has been provided. The work submitted is the result of my own investigations conducted under the supervision of Professor Richard Hastings.

For chapter two I was provided with support at the analysis stage and feedback on the manuscript by Dr Vaso Totsika and Dr Emma Langley. This was conducted during the preparation of this thesis and has been published prior to submission: Hayden, N. K., Hastings, R. P., Totsika, V. & Langley, E (2019a) A population-based study of the behavioral and emotional adjustment of older siblings of children with and without Intellectual Disability. *Journal of Abnormal Child Psychology*. 47, 8. 1409-1419. doi: 10.1007/s10802-018-00510-5.

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Summary

Siblings of children and adults with intellectual and developmental disabilities (IDD) have been the focus of research attention for several decades. This PhD thesis sought to build on that knowledge base, and included data about over 1,000 siblings of adults and children with IDD. This thesis has been able to draw upon large scale survey data to further understand siblings' psychological outcomes as well as exploring areas less frequently considered in the sibling literature, including siblings' school experiences and sibling relationships. Chapter one consisted of three main sections: a discussion about conceptual and theoretical perspectives about families, an overview of relevant, methodologically robust empirical literature, and a self-reflexive section that explored my experiences as a sibling. These three sections each highlighted methodological issues related to existing sibling research. Chapter two used representative, population-level data from the Millennium Cohort Study to examine the behavioural and emotional outcomes of older siblings of children with ID ($n = 257$) in comparison to older siblings of children without ID ($n = 7246$). Chapter three reported on an initial evaluation of a school-based intervention (Sibs Talk) for siblings ($N = 55$). Chapter four provided findings from the Adult Sibling Survey, these data constitute the largest UK sample of adult siblings of people with IDD ($N = 911$). This chapter examined adult siblings' mental distress, wellbeing, quality of life, and health outcomes, using moderated multiple regression analyses. Chapter five presented sibling data from the 1,000 Families study. This study drew upon structural equation modelling to examine reciprocal effects about sibling relationship outcomes by considering the behavioural and emotional outcomes of both siblings ($N = 500$ sibling dyads). Chapter six synthesised the main contributions of these four empirical chapters, including a discussion about methodological limitations, as well as research, practice, and theoretical implications. Cumulatively, the research reported in this thesis showed that child and adult siblings had poorer psychological outcomes in comparison to other samples, but these poorer outcomes were likely related to contextual factors, such as socio-economic status and caring roles. Sibling dyad level analyses supported conceptualisations of reciprocity in the family system, with both children's behaviour associated with sibling relationship quality. Sibs Talk was associated with promising positive outcomes. Notably, a simple negative narrative about the effect that people with IDD have on their non-disabled siblings was not supported by the evidence in this thesis.

Chapter 1: Introduction

Context and Definitions

An intellectual disability (ID) is defined as a disability that affects an individual's adaptive behaviour as well as their intellectual functioning that has an onset before the individual is 18 years of age (AAIDD, 2019). A meta-analysis of international prevalence studies indicated that the prevalence rate for ID is approximately 1% (10.37/1000 population, 95% CI 9.55, 11.18 per 1000 population) of the global population (Maulik et al., 2011). A study using USA-based population data indicated this figure could be higher, estimating that the ID prevalence rate was 3.2% (Platt et al., 2019). Developmental disabilities (DD) include disabilities such as autism, Down syndrome, Fragile X syndrome, Williams syndrome, Cerebral palsy, or (for young children) global developmental delay. For some DDs such as Down syndrome, ID is an integral part of the condition. For other DDs, such as autism or Cerebral palsy, ID may or may not be present.

The past few decades has seen the development of research about families of people with intellectual and developmental disabilities (IDD). Empirical research suggests parents and siblings of people with IDD may be at risk of worse psychological outcomes than other parents and siblings (Singer & Floyd, 2006; Emerson et al., 2010; Marquis et al., 2019). For siblings more specifically, research often positions siblings as a solution to future caregiving needs for people with IDD (Leane, 2019), with siblings potentially caring for their brothers and sisters with IDD when their parents are no longer able to. People with IDD are living longer lives than ever before (Coppus, 2013). Improvements in health and life outcomes for disabled people (e.g. increased life expectancy amongst people with Down syndrome; Hodapp & Urbano, 2007), along with the reduction in state welfare available in various national contexts, has prompted questions surrounding the uncertainty of care and support for people with IDD (Power & Bartlett, 2019). Therefore, if we care about the outcomes and the quality of life of people with IDD, then we must also consider the outcomes of their brothers and sisters without disabilities. If we are interested in siblings' caring potential, then we should also be interested in understanding and fostering sibling relationships, as this has been associated with a willingness to take on this caring or supporting role in the future (Burke et al., 2012).

A family is characterized, in essence, by relationships between a group of individuals that holds socio-cultural-legal significance. Although a thorough exploration of the family is beyond the scope of this thesis, I will attempt to provide a working definition of both the family and the sibling relationship. Raymond Williams (1976) summarized the significance of the concept of the family through social history and traced etymological usage of the term. Earlier conceptualizations of the family, such as the household (referring to a dwelling where relatives and servants resided together), familiar (referring to a range of relationships but in essence indicated a closeness or familiarity without a genetic relationship), or references to the family in religions (e.g. the father, the son, the holy mother, brothers and sisters), were essentially referring to large kin-groups. Williams (1976) indicates that the more familiar conceptualization of the family as a small kin-group emerged as dominant in the Western context between the 17th to the 19th centuries before dominating in the 20th century. This highlights to us that the ‘traditional’ nuclear family, consisting of a mother, a father, and children, is a relatively modern social structure. Therefore, it is important to emphasize that the concept of the family is not universal or fixed, it is socio-culturally constructed and perpetually changing. For example, the latter half of the 20th century saw medical advances such as the introduction of in-vitro fertilization as well as increases in divorce rates, leading to single-parent households and step-families becoming more common. The early 21st century has seen the recognition of LGBTQ+ people’s rights to marriage, adoption, and access to medical fertility options. There is not and there has never been a ‘traditional’ family and therefore, we need to think openly and flexibly about how we define the family. Rosenblatt (1994) provided the following definition:

‘...the essential character of the family entity includes culturally defined family roles and role interactions that differ by age and gender. It includes culturally defined family identity markers as, for example, a name, a dwelling, genealogical relations, commitments made by members to each other, or legal rights and obligations to one another’ (Rosenblatt, 1994: 35).

This definition is useful because it is relatively flexible and open. It emphasizes that the family is culturally and socially defined, it allows for the inclusion of extended family members as well as immediate family members to be included, and it does not only prioritize, for example, genetic relationships or those that reside with one

another. The simplest definition of siblings would be people who share one or two parents. Sanders (2004) highlights that this may exclude various sibling relationships, such as step, adopted, or foster siblings, as well as children that are raised together but share no genetic or socio-legal relationship. Reading Rosenblatt's definition as quoted above to understand sibling relationships would allow us to incorporate these non-genetic sibling ties, maintaining an open and inclusive definition of both the family and the sibling relationship.

The following three sections explore some of the issues and assumptions in the existing field by reviewing three distinct areas: conceptual and theoretical perspectives, existing empirical sibling research, and self-reflexive perspectives. At the end of each of these three sections, I summarize some of the methodological and ethical issues raised by each area before I provide an overview of the remaining thesis chapters.

1. Conceptualizing Families and Siblings

Family Systems Perspective

The most common framework for conceptualizing siblings in the IDD family literature is arguably family systems perspective (Cridland, 2014). The application of systems thinking to family studies was initially advanced by Murray Bowen (1966) who argued that understanding individuals as part of a family, and the family as a system, may help us move beyond static conceptualizations of people and families (Bowen, 1966). The basic premise for Bowen, was that 'a change in one part of the system is followed by compensatory change in other parts of the system' (Bowen, 1966: 351). He described the way in which one family member 'over functions to compensate for the dysfunction [sic] of the other' (Bowen, 1966: 351) and that if the conditions are ongoing, this can lead to a lack of flexibility and reduced functioning within the family system. Family systems is, in essence, a way of understanding the way in which a person within a family system can have an effect on other people within the family system 'wherein individual family members are necessarily interdependent, exerting a continuous and reciprocal influence on one another' (Cox & Paley, 1997: 246). Something that is at times lost in the IDD sibling research is the reciprocal influence siblings and family members can have on one another.

It is noteworthy that in the wider sibling literature, social learning theory is the most commonly applied theory (Feinberg et al., 2012; Whiteman et al., 2011), and it is valuable to question why this model is underutilized in the IDD sibling literature. This perspective considers the ways in which siblings learn and develop from one another, acquiring behaviours, attitudes, and beliefs (Whiteman et al., 2011). Evidently, family systems perspective also allows for this sort of modelling through the assumption of reciprocity. It is important therefore that the examination of bidirectional and reciprocal effects are not lost in IDD sibling research. An issue with the way in which a family systems perspective is often applied in the IDD literature is that it has been used to justify a more negative narrative about siblings' outcomes and experiences. This is illustrated in the psychological outcomes literature about siblings, whereby the design and conceptualization of these studies rest on the assumption that the sibling is the target and receiver of research attention due to the effect we assume having a brother or sister with IDD may have on the sibling. Hastings (2016) argues there is a case for rejecting the negative narrative often premised in IDD family research. Hastings traced empirical data about families of children with IDD to highlight that families do not universally experience 'negative' outcomes, and some studies indicate 'positive' outcomes for families. Hastings (2016) suggests that further systems-based analyses and questions may provide an appropriate framework for disrupting the negative narrative about families of people with IDD, such as by exploring reciprocal effects, rather than focusing on the one-way effect that the person with IDD has on each family member.

A deeper overview of family systems perspective is therefore necessary to understand how it can help us re-conceptualise families and sibling dyads. According to White and Klein (2002), family systems rests on the following four principles: firstly, that every element of the family system is interconnected. Secondly, only by considering the whole family system, can individuals within the family be understood (i.e. a 'meaningful whole'; Smith-Acuña, 2011: 6). Thirdly, there is 'feedback' within the system, or reciprocal effects (i.e. 'necessarily interdependent...and reciprocal influence'; Cox & Paley, 1997: 246); and lastly, the theory should be understood merely as a 'way of knowing' (White & Klein, 2002: 123) rather than as a reality i.e. reification should be avoided and family systems perspective should be used heuristically to help us understand families further. This

fuller definition of family systems emphasizes the potential of family systems perspectives to move beyond simple effects that are premised on the assumption that family members will be negatively affected by having a family member with an IDD.

For Turnbull and Turnbull (2001), they define three main assumptions related to family systems perspectives, firstly, they describe *inputs and outputs* related to the interactions between family members and the roles the family members perform. Secondly, they emphasize the *wholeness and subsystems* within the family system. Thirdly, and most distinct from the description provided by White and Klein (2002) is the assumption of *boundaries*. Boundaries are formed by interactions between family members and outside interactions that define, or create norms, for behaviours, roles, and expectations between members of the family, including those outside of the household. Boundary ambiguity occurs when these boundaries are more permeable (Cridland et al., 2014). This may be particularly salient for families of people with IDD, where these boundaries may at times become blurred. More frequent changes in the system, such as in behaviours perceived as challenging or the involvement of external services, may result in some siblings taking on more household tasks or caring responsibilities than would usually be expected (Cridland et al., 2014). Ideally, boundaries within subsystems are flexible, and therefore more able to be influenced, although not determined, by other elements of the family system (Whiteman et al., 2011). This concept of boundaries also has implications for practitioners and researchers. Families vary in how open their boundaries are to outsiders (Turnbull & Turnbull, 2001). For researchers, the data we are able to collect from family members may be defined and restricted by this insider-outsider boundary.

The Sibling Subsystem

Family systems are understood to be structured and organised hierarchically (Smith-Acuña, 2011) with subsystems embedded within the overarching family system ‘that are really systems of their own’ (Cox & Paley, 1997: 245). Subsystems may include the couple/parental subsystem, the parent/child subsystem, and the sibling subsystem – along with subsystems involving extended family members and grandparents (Dallos & Draper, 2015). Only by understanding the subsystems can

the wider family system be understood, and *vice versa* as ‘any individual family member is inextricably embedded in the larger family system and can never be fully understood independent of the context of that system’ (Cox & Paley, 1997: 246).

Therefore, a focus on siblings and the sibling subsystem has value from a family systems perspective. This also highlights the reciprocal nature of the effects. Much of the empirical research about siblings focuses on the outcomes of the non-disabled sibling (Cridland et al., 2014), which conceals the emphasis family systems theory puts on the reciprocal effects i.e. that the non-disabled sibling has on their brother or sister with IDD and their parents. The way in which these different subsystems interact is complex (Cox & Paley, 1997). Subsystems and dyadic modelling of sibling relationships have been criticised for oversimplifying the complex and overlapping nature of sibling and family dynamics (Sanders, 2004). This is because the interfamilial relational pathways are more than simply the association between the different individuals within a family, but also the influence between the relationships and the individuals, as well as the influence of these relationships with other relationships.

To add to this complexity, these influences and dynamics are constantly changing. Therefore, Sanders (2004) calls for us to ‘adopt a position of humility in the face of the immense complexity of this social system called a “family”’ (Sanders, 2004: 33). The inter-related and reciprocal nature of these subsystems is important in understanding the overall family as well as each aspect of it (Cox & Paley, 1997; Cox, 2010). The hierarchical aspect of the way family systems are conceptually organised does emphasise the importance of the parent-child relationship, potentially neglecting other aspects of the system, such as the sibling subsystem (Cox, 2010). Rosenblatt (1994) highlights the root of ‘sub’ as less than or secondary to something else – i.e. the wider family system. If we consider the uniquely long-lasting nature of a sibling relationship, this minimizes the importance of sibling relationships.

Taken cumulatively, at the sibling dyad level, we must consider not only how each sibling effects the other, but also how the dyad as a whole functions to understand the sibling dyad, and the sibling as an individual. It is worth re-emphasising that subsystems, as with the overall family system, have ‘heuristic value but no “ontological” reality’ (White & Klein, 2002: 127).

Embedded Systems Thinking

A criticism of family systems perspective is that it can be seen to isolate a family from their wider context and the larger systems in which the family is embedded (Rosenblatt, 1994). However, family systems are open systems (Whiteman et al., 2011) and are therefore affected by external influences. Cridland et al., (2014) explored systems perspectives in relation to families of autistic people. Cridland et al., (2014) divided family systems perspectives into broadly two categories: *macroscopic* approaches and *microscopic* approaches to family systems. Macroscopic approaches consider the way that the family system interacts with other systems external to it, such as schools or community contexts. Microscopic approaches consider within-family factors, such as focusing on subsystem analyses. Both approaches are important in helping us understand the family system, but it is important to also consider the way these factors interrelate (Cridland et al., 2014).

Some work has been done to further incorporate elements from the macrosystem, exosystem, mesosystem as well as microsystem (*cf.* Bronfenbrenner's ecological model; 1979) into a more comprehensive model about siblings of autistic children (*cf.* 'Embedded Systems Framework'; Kovshoff et al, 2017). Kovshoff et al's (2017) approach helps us to consider the multiple, interacting factors at play in understanding the experiences of siblings who have a brother or sister with a disability. A full range of factors were incorporated into the framework such as: the influence of peers, school, social media, social class, culture and ideology. This enables us to further our understanding of the wider and more interactive factors related to siblings' psychological, social, familial, and educational outcomes as well as relationships. In a more practical way, this model helps us to consider what covariates we might include in our statistical models. It also highlights the need for analyses that move beyond simple main effects, and to consider how these different factors may interact and overlap with siblings' experiences. Although the 'Embedded Systems Framework' was about siblings of autistic people, I consider that it advances our understanding of siblings of people with other disabilities as well, such as ID and other DDs.

Dynamic and Fluid Systems

What is most interesting about family systems perspective, as highlighted by the four aspects defined by White and Klein above (2002), is the conceptualization of movement and exchange between members of the family. It highlights the need to move beyond psychological and individualised approaches, to not merely think about the individual elements of the family system, but to consider the ways in which these elements interact with one another (Rosenblatt, 1994). Therefore, family systems perspectives justify exploring sibling relationships as well as considering the outcomes of children with IDD and their siblings as individuals. For Rosenblatt (1994), family systems perspective has the potential to help us understand families beyond the sum of each part of the system, or each individual family member, but this can only be done by also exploring the way each part of the system interacts, such as with sibling dyad-level analyses.

A family systems perspective might appear to be rather static, and not dynamic enough for social, family research, where change is constant. Some models have been called 'structure and process models' in order to capture the way in which the family system is unstable (Buckley, 1967). Cox and Paley (1997: 245) apply general systems theory to further capture the dynamic potential of family systems. They describe family systems as having homeostatic features, compensating 'for changing conditions in the environment by making coordinated changes in the internal workings of the system' and having 'the ability of open, living systems to adapt to change in or challenge to the existing system' (Cox & Paley, 1997: 245). This is helpful for conceptualising families as the changeable and unpredictable entities that they are. Although family systems are described as dynamic, the nature of modelling such an ambivalent and unstable concept, as the family is, obscures this. Rosenblatt argues that 'despite the notion of family systems as dynamic, the system metaphor obscures the fluidity of family composition' (Rosenblatt, 1994: 55). This may be due to the nature of drawing out a family system – the fluidity and dynamic-ness of the system is lost because committing anything to the page makes it somewhat static and permanent. Family relationships, particularly siblings' relationships are very changeable and dynamic. Any theory attempting to model 'the family' as a complex psychosocial construct will inevitably oversimplify the concept. Inadvertently therefore, any statistical analysis or modelling of questions related to sibling relationships or experiences will inevitably over-simplify them.

This is not just a matter related to quantitative research and family systems perspective; it arguably permeates all social research. For Rosenblatt (1994), they describe this as:

‘...fluidity arising from the multiple ways in which the reality of the moment can be perceived and characterized. Yes, the system is dynamic, but perhaps what seems like dynamics arise in part from our limited capacity to know or make sense of what is going on at any given moment’ (Rosenblatt, 1994: 55).

Family systems perspectives can in this way be seen as not just a tool to model and contextualize families’ outcomes and experiences, but also as an epistemological framework to consider the limitations of conducting sibling and family research. I believe that this is not however, a problem only for family systems perspectives. This limit in our ‘capacity to know’ (*Ibid.*) permeates all social research. Family systems perspectives are therefore useful for precisely the reason that they are limited. By defining, simplifying and essentializing a process, we can never adequately capture the phenomena itself. In this way, it mirrors the research process – whereby we collect specific data from our participants at specific points in time, cognizant that the moment we do, and certainly by the time we begin our analyses, their perceptions about their experiences and outcomes would have changed and therefore, our interpretations of their perceptions are already outdated. This is not to say that our data and findings are not useful and valuable, but that any dissatisfaction with one theory, or one broad methodological approach (i.e. quantitative vs. qualitative research) can surely be levelled at another. In this way, we need to adopt a flexible and pragmatic approach to the theories, methods, and approaches that we use.

A common question asked about family systems is the extent to which it is a ‘theory’ or whether it is a ‘model’ (White & Klein, 2002). Systems thinking can be used as an overarching theory to consider siblings as the subject of research attention and potentially as recipients of supports and interventions, because we could use family systems to explain that siblings may experience different outcomes, such as being at risk of worse outcomes, because they have a brother or sister with IDD. These perspectives can help us explain, conceptualize and interpret our research in broad ways. However, no theory or model can fully capture the reality of socialised

experiences – and so we will always question to what extent a social hypothesis is testable. Whenever any social idea is thought, spoken, modelled or written about, something is lost, the idea or model almost fossilises. The idea entombs reality and the reality ceases to exist. A more important question is; is the idea *useful*? Does it help us understand phenomena further? Does it resonate across families? Does it help us conceptualize and design our research better? Is it dynamic enough to account for differences and changes both within and between families? Does the concept help us to support and improve the lives of the people and the families we work with? Family systems perspective may not be a particularly beautiful concept, but it is useful. Another criticism of family systems perspective is that it is so broad and general that it is meaningless (White & Klein, 2002). However, its generality provides its flexibility and therein lies its usefulness and utility for researchers.

Lifespan and Developmental Perspectives

To further consider the importance of understanding change within the family system, it is useful to consider family life course perspectives and family developmental perspectives. Some writers have incorporated developmental frameworks with family systems perspectives to explore, for example, anticipatory loss related to disability (Rolland, 1990). These ideas help us to consider how families change over time through stages and events in the family life course. For White and Klein (2002), these perspectives are conjoined – or perhaps conflated – into a single framework: ‘The Family Life Course Developmental Framework’. These perspectives consider the ways in which families are embedded within a kinship structure, with changing family positions, roles and norms.

For families with a family member with IDD, siblings may provide more support and care for their brothers and sisters with IDD than in other families, blurring some of the distinctions of these roles and family positions. This concept overlaps with the focus on boundaries in the family systems framework. Siblings are often expected to care and ‘watch out’ for one another, particularly older siblings for younger siblings (White & Klein, 2002). For siblings with a brother or sister with IDD, these norms may differ, as they may provide more support than would usually be expected, and younger siblings without a disability may provide care and support for their older brother or sister with IDD. Societal norms expect siblings to provide

friendship and support for one another in adulthood but for sibling dyads where one has an IDD, these may appear less reciprocal or balanced than for other sibling dyads.

Developmental perspectives suggest that sibling relationships become less salient in early adulthood, and more important again in later life (White, 2001). Family life course and developmental perspectives also focus on family stage and transitions between these stages. For siblings where one has an IDD, their relationship may not follow this pattern. Some work has been done to explore transitions from childhood to adulthood for siblings of people with ID or autism (Todd, 2019) and consider how sibling relationships and experiences when one sibling has a disability change over time. Transition periods for families of a child with IDD may include 'typical' transitions, such as biological and social changes in adolescence. Transitions may also be different, such as the transition from childhood to adulthood leading to a disruption in support, as the child with a disability moves from child to adult services. There may also be additional transitions for the families of a disabled person, such as during the diagnosis process, or during developmental milestones for the person with IDD (Cridland et al., 2014) which may be more spaced out, and potentially perceived as more significant by the family.

These perspectives highlight the importance of understanding siblings' relationships, outcomes and experiences throughout the life course, rather than focusing predominately on, for example, childhood and later adulthood (White, 2001). Reading developmental and lifespan perspectives in relation to family systems perspectives helps us further understand how families change over time.

Double ABCX model

The Double ABCX model (Hill, 1949; Hill, 1958; McCubbin & Patterson, 1983) considers how the family system adapts in response to a situation, or stressor. The ABCX model is conceptualised as: the *A factor*, the stressor event and hardships; the *B factor*, the family's resources to respond to the stressor, such as family integration and family adaptability; the *C factor*, which is the focus the family have on the stressor and their perceptions and outlook of the event; and the X factor, how factors A, B, and C interact within the family system to produce, or prevent, a crisis. The Double ABCX model was developed by McCubbin & Patterson (1983) to

expand the ABCX model to consider the dynamic processes and cumulative stressors that families experience:

aA considers the pile-up of stressors or demands, and these may stem from an individual, the family system, or the wider community or society that the family is embedded in. If we apply this to families of a disabled person, the initial situation would be the birth or the revelation that their child has a disability (i.e. the diagnosis process). Stressors may pile up, for example, one parent may decide not to work to provide extra support to their child, and this may lead to a reduction in financial resources and stresses related to loss of income.

bB considers the resources families apply in response to the situation. This could be existing resources, such as being part of a religious community, financial resources, or having the social and cultural capital to effectively navigate support services for a disabled family member. It could also be resources that the family acquired in order to respond to the situation. So this could be by accessing community-based support groups for parents of disabled children, developing skills to more effectively navigate support services, or applying for extra support from the local authority for educational or home adaptations and adjustments.

cC describes the changes families make to their interpretation of the situation or stressor. Families may find the initial diagnosis upsetting, or they may find it affirming of the differences they have observed in their child, and a tool for getting their child the support they need. For families of disabled children their perceptions may oscillate. For example, a parent who has spent the afternoon observing and comparing their child to other children of a similar chronological age may interpret their situation less favourably than a parent who has just witnessed their child reach a developmental milestone or display prosocial behaviours towards them.

xX describes the way the family adapts to the stressor. For families of a disabled child, this considers whether there is a balance between demands and capabilities. For example, that the demands of the child are met by the capabilities of the family, and that the demands of the family system are met

by the wider community or societal context. It also considers how the coherence of the family, along with the balance between demands and capabilities, lead to the outcome. The outcome is understood as a continuum (i.e. a continuous variable), that is expected to constantly fluctuate due to the changes and processes occurring as described above (i.e. aA, bB, and cC).

Applying this model to statistical analyses, the effects between aA, bB, and cC can be conceptualised as direct effects to xX; bB, and cC could be conceptualised as mediating variables between aA and xX (Saloviita et al., 2003); and the interaction between bB and cC can be seen as moderating effects (i.e. BC; Meleady et al., 2020). In this way, the model can not only help us consider which variables we should include in our analyses as covariates, but also consider analyses that move beyond direct effects, e.g. by considering moderating effects. The model has been used to understand the experiences of families with children with autism (McStay et al., 2014; Bristol, 1987; Pakenham, 2005; Meleady et al., 2020), as well as parents of children with ID (Saloviita et al., 2003). The model is useful because it helps us conceptualise why some families and siblings of people with disabilities may have better outcomes, or be more ‘resilient’ than others. The extended ABCX model provokes consideration of multi-level and contextual factors in our analyses, as it incorporates individual-level, family-level, and societal-level factors. These levels can be seen to overlap with extensions of systems thinking already discussed, such as Bronfenbrenner’s macro, exo, meso, and microsystems (Bronfenbrenner, 1979) and the Embedded Systems Framework (Kovshoff et al., 2017). This model therefore indicates that stress is not uniform. We should expect to see siblings and families of disabled people responding differently from one another. McCubbin and Patterson (1983) also introduced the ‘Family Adjustment and Adaptation Response’ (FAAR; Patterson, 1988) model which brings in further consideration of changes over time and a phased approach to considering family adjustment and adaptation. These models can therefore help us further consider the non-static nature of the family system. FAAR is also seen as a turning point in the conceptualisation of family stress as a move away from family weaknesses towards considerations of family strengths and resilience (White et al., 2015), therefore, helping us to move beyond an assumed negative narrative about sibling and family experiences.

Summary of Theoretical Approaches

In summary, family systems perspectives have commonly been used in IDD sibling research as a legitimising tool for focusing on siblings, by suggesting that the person with IDD may be having some sort of ‘negative’ effect on the sibling. This is still an important area to understand, but sibling research must move beyond these types of analyses as well. As explored in this section, family systems perspectives also enable us to conceptualise more interesting analyses than these direct effects, such as reciprocal effects at the sibling dyad level. Embedded models and models that explore effects outside of the family system help us consider which variables to include in our analyses and reinforce how vast and unknowable individual and family experiences are. They help us consider that communities external to the family system (e.g. schools) may be associated with siblings’ outcomes and relationships.

The Double ABCX model helps us understand family stress and resilience when a child in the family has IDD, further mapping variables we could include in our models, such as the effect of socio-economic factors. It also encourages us to move beyond direct effects where possible by, for example, exploring moderating effects. The life span and developmental perspectives briefly introduced, highlighted the importance of understanding change and transition, but also justifies exploring the outcomes of child and adult siblings as interconnected rather than separate entities. Although the concept of time is often included in all of these models, this element of change within the family system also highlights an issue with these models and research generally, and that is the transient and heuristic nature of capturing any perspective, about a theory or model, or indeed data from a research participant.

2. Review of Existing Literature

The theoretical perspectives just discussed can be used to contextualise the assumptions made in the existing empirical literature about siblings. Empirical research about siblings of people with disabilities has tended to focus on psychological outcomes, particularly for child siblings. This stems from the way family systems theory intersects with a negative narrative, whereby siblings are assumed to be negatively affected in some way due to having a brother or sister with a disability. This section will focus on reviewing the existing empirical research

about the outcomes of both adult and child siblings. The most consistent point made in existing sibling studies is that findings about siblings' outcomes are mixed and inconsistent. For example, some studies have found that siblings have worse outcomes (Hastings, 2003; Verté et al., 2003; Ross & Cuskelly, 2006; Orsmond & Seltzer, 2007; Goudie et al., 2013) and some studies have found no evidence of differences in outcomes (Cuskelly & Gunn, 2006; Hastings, 2007; Howlin et al., 2015). These mixed findings are likely due, at least in part, to methodological issues, such as an over-reliance on small, convenience-based samples that were biased. Rather than provide a comprehensive literature review here and risk over-emphasising small-scale studies, I will focus this overview of the existing literature on methodologically strong studies, such as population based-data and systematic reviews where available. Where not available, I will draw upon large-scale and longitudinal studies with 'normative' comparison samples. This may mean drawing on some non-IDD sibling literature where methodologically appropriate.

Outcomes of Child Siblings

The existing empirical literature about the outcomes of child siblings of children with disabilities has included an early systematic review and meta-analysis summarising the psychological outcomes of siblings with ID (Rossiter & Sharpe, 2001), drawing on 25 studies with 79 distinct effect sizes. Although the inclusion criteria included both adult and child sibling studies, there was an over-representation of child sibling studies with only two of these 25 studies including adult-only samples. Overall, they found that there was a small negative effect for siblings of people with ID. Dependent variables included measures of: psychological functioning, self-concept, externalising and internalising behaviours, depression, and anxiety. Rossiter and Sharpe (2001) highlighted that the negative assumption may be overstated and that the small negative difference they did find may have been due to methodological and publishing biases. Sharpe and Rossiter (2002) also went on to conduct a meta-analysis about siblings of children with chronic illness (51 studies, 103 effect sizes) and found again a small, negative effect size for siblings of children with chronic illness in comparison to other children. Again, they suggested that more robust methodological studies are required to understand if the difference was meaningful.

Over the last decade, population-based studies about the psychological outcomes of siblings of children with a disability (Neely-Barnes & Graff, 2011, Goudie et al., 2013, Emerson & Giallo, 2014) and about siblings of children with DD more specifically (Marquis et al., 2019, Marquis et al., 2020) have been published. Population-based data provide large-scale, representative data about a given sample. This limits the effects of sampling biases on resulting analyses. Goudie et al. (2013) used data from the USA Medical Expenditure Panel Survey and they found that siblings of children with a disability were 2.77 times more likely to have significant functional impairment, were more likely to have problems in school, and have more social problems compared to siblings of children without disability. Neely-Barnes and Graff (2011), using USA national health data, found that differences between siblings of children with disabilities compared to siblings of children without disabilities were non-significant once other predictors were controlled for, such as socio-economic status (SES). Emerson and Giallo (2014) had similar findings. Using nationally representative data from Australia, they found that siblings of children with disabilities had worse wellbeing compared to other siblings for some adjustment areas, although not all adjustment areas. They found that once they controlled for SES, these group differences were non-significant. The findings from Neely-Barnes and Graff (2011) and Emerson and Giallo (2014) highlight the importance of considering wider contextual factors beyond the immediate family system in order to understand sibling outcomes (Kovshoff et al., 2017). In particular, variables related to SES were important factors in understanding siblings' outcomes. Population-data in the wider IDD literature tells us that families with a child with IDD were more likely to experience lower SES (Emerson, 2003; Emerson, 2004). Therefore, the importance of SES should not be underestimated in IDD family and sibling studies.

Recent work using Canadian population-level administrative data has explored the outcomes of siblings of children with DD (Marquis et al., 2019, Marquis et al., 2020). Marquis et al. (2019) used Canadian population data from the Ministry of Health in British Columbia. These data benefited from deriving mental health information about siblings as measured by health services rather than self-report. They found that siblings of children with DD had increased odds of depression and other mental health problems even when other variables were

considered. Therefore, although important factors such as SES were also associated with siblings' outcomes, having a brother or sister with DD was still associated with siblings' outcomes even once these factors were considered. In Marquis et al., (2020) these analyses were extended to examine the outcomes of siblings of children with four different DDs: autism, foetal alcohol syndrome (FAS), Down syndrome and an 'other' group. Three models compared siblings of children with autism, FAS and 'other' DD, to siblings of children with Down syndrome. The variable was statistically significant in each model – indicating that siblings of children with Down syndrome had better mental health outcomes compared to the other three sibling groups.

In terms of other methodologically robust research about child siblings, there have also been three systematic literature reviews exploring interventions for siblings of children with disabilities and chronic health conditions (Hartling et al., 2014; Tudor & Lerner, 2015; McKenzie Smith et al., 2018). These reviews highlight how most interventions for siblings of children with disabilities are group-based and focus on providing supports in family, medical and community contexts e.g. summer schools and community sibling groups. There may be further scope to explore whether schools may also be able to provide additional support for child siblings. In terms of school-based outcomes for siblings of children with disabilities, the population data from Goudie et al., (2013) found that siblings were more likely to have functioning problems with behaviour at school, and have problems with completing their school work compared to siblings of children without disabilities. A systematic review about the school-based experiences of siblings of children with chronic illness (Gan et al., 2017) identified 28 eligible studies and found a focus on the psychological impact, problems with attendance and academic functioning, and difficulties with interacting with peers and teachers. These data further support the potential of a school-based intervention for siblings.

Outcomes of Adult Siblings

The most robust data about adult siblings were reported by Rai et al. (2018). Rai et al. (2018) used a Swedish population-based cohort study with 223,842 participants to explore depression in young adults aged between 18 and 27 years old (mean age 21.5), comparing depression in young adults with and without autism. As

a supplementary question in order to explore genetic effects, they also reported and compared depression with their full and half siblings. They also included sociodemographic factors in their analyses. The study reports on both siblings of people with autism plus ID and siblings of people with autism without ID. The age of the siblings is not reported as it was merely a secondary research question in this study, but we assume the adult siblings were also young adults – as the depression measure reported was for adults and most of the siblings were likely to have been a similar age to their brothers and sisters within the sample. Rai et al. (2018) found that both full and half siblings of young adults with autism were slightly more at risk of having a depression diagnosis than the control group/general population and this was even the case when they controlled for age, sex, family income and other commonly considered effects in adult sibling research.

Another large-scale study exploring siblings of adults with disabilities in comparison to normative samples is the Wisconsin Longitudinal Study (WLS). The study is a random-sample, longitudinal study that follows the lives of a group of adults ($N = 10,317$) who graduated in 1957 from high schools in Wisconsin, USA (Herd et al., 2014) throughout their life-courses. Selected siblings were incorporated into the study in 1977, 1994, 2005 and 2011. These data have provided the foundation for a number of analyses exploring group differences for adult siblings of people with particular disabilities in comparison to adult siblings of people without disabilities. Drawing on data from WLS, Taylor et al. (2008) found that adult siblings of people with mild ID had similar scores on the psychological wellbeing and distress measures as well as in the personality measures when compared to the sample of adult siblings of people without a disability. Adult siblings of people with mental illness however, were more likely to have had a depressive episode in the past, scored higher on neuroticism, and scored lower than the group of siblings of people without a disability on the conscientiousness, extraversion, psychological wellbeing, and agreeableness scores.

Both Rai et al. (2018) and Taylor et al. (2008) report on data from within a specific geographical location within a country. Other studies have benefited from recruiting large sample sizes across a wider, national area. Hodapp et al. (2010) used a national, online survey to explore the differences for male and female adult siblings of people with disabilities. There were 1,160 adult siblings in the sample that

included measures to explore a range of areas, including adult siblings' emotional and physical wellbeing. Hodapp et al. (2010) did not have a normative sample to compare the outcomes of adult siblings, so instead compared male and female adult siblings of individuals with disabilities. They suggest male siblings may report slightly fewer physical and mental health issues than women, but overall, most siblings reported good physical and mental health – although there was no control group to compare the results to. Hodapp and Urbano (2007), using a sub-sample of the same collection of data, compared the self-perceived health and depressive symptoms as well as the relationships of adult siblings of people with autism ($n = 176$) in comparison to siblings of people with Down syndrome ($n = 284$). They identified small, worse outcomes for the adult sibling group of people with autism compared to siblings of people with Down syndrome. These studies, although useful, highlight the need for comparative data, via recruiting a 'normative' sample or by using measures with population 'norms' available.

Sibling Carers

There are some methodologically robust studies about adult sibling carers. Sonik et al. (2016) used longitudinal USA population-level data from the Survey of Income and Program Participation to describe the demographic characteristics of sibling carers of people with IDD and compared them to the general working-age population. They found that moderate material hardship was more common amongst sibling carers. They also found that sibling carers were more likely to be older, women, Black and less likely to have ever been married. Lee and Burke (2018) have conducted a systematic review to explore carer roles of adult siblings of people with IDD. They summarised that adult sibling carers reported benefits as well as challenges related to caring for their brother or sister with IDD and that the predictors of future caring were inconsistent across different studies.

Methodologically, Lee and Burke (2018) criticized the lack of a consistent definition of caregiving across the sibling carer literature. Larger-scale adult sibling surveys have been conducted in the USA as already cited (Hodapp et al., 2010; Hodapp & Urbano, 2007). Subsets of this large sample have also been used to understand sibling carers specifically, such as by predicting future caregiving expectations (Burke et al., 2012). More recently, members of this research team have conducted another survey in collaboration with the Sibling Leadership Network, USA, and

again, subset studies were used to explore the association between, for example, disability severity and sibling caregiving (Lee et al., 2019a). These surveys are large-scale, having recruited more than 1,000 participants in the over-arching studies. For child carers, there are fewer studies about young carers generally, and these tend to be methodologically limited. We have not identified any specific studies about child sibling carers. The present caregiving literature does help us understand which siblings may be more likely to provide care, but does not help us understand the outcomes and therefore the potential support needs of siblings that are also carers.

Sibling Relationships

As mentioned, a great deal of the research attention about siblings has focused on psychological outcomes. However, less is known about sibling relationships in the IDD literature, and I have identified no population-based, large-scale data about sibling relationships, nor systematic reviews. This previous focus on the psychological outcomes of the non-disabled sibling has been problematized as leading to the marginalization of disabled people and leading to more limited sibling research (Meltzer and Kramer, 2016). Sibling relationships are important for both siblings, and will likely be the longest relationship many people have in their life time, having the potential to last across the life-span, therefore ‘siblings have a significant capacity to influence each other’s lives’ (Meltzer and Kramer, 2016: 18). Furthermore, sibling relationships enable children to learn social behaviours that then influence relationships siblings have outside the family home (Mandleco & Mason Webb, 2015). Existing statistical analyses about child sibling relationships often encompasses measures of warmth, closeness, conflict, status/power and rivalry (Furman & Buhrmester, 1985). In adulthood, there are questionnaires measuring affect, behaviour, and cognitions in adulthood as well as retrospective reflections about these dimensions in childhood (Riggio, 2000). There are also adult sibling relationship questionnaires measuring warmth, conflict and rivalry (Lanthier & Stocker, 1992). These measures were developed in the wider sibling literature, where neither sibling has a disability. More recently, a measure has been developed for non-disabled adult siblings to answer questions about their relationship with their disabled brother or sister (Sommantico et al., 2020) capturing five dimensions: closeness, conflict, jealousy, self-marginalization, and worry.

Studies exploring sibling warmth and closeness when one has a disability have found particularly ‘positive’ sibling relationships for example, when one sibling has Down syndrome compared to when one sibling has autism (Hodapp & Urbano, 2007). Although many researchers have hypothesized that sibling relationships where one child has an ID might have more negative relationships, the empirical evidence base for this hypothesis is currently unsubstantiated. Existing research exploring more ‘negative’ aspects of the sibling relationship have found similar levels of conflict between typically developing sibling pairs compared to sibling pairs where one has an ID (Doody et al., 2010). Other studies have found that sibling pairs where one has an ID may have less conflict in their sibling relationships than other siblings (Floyd et al., 2009; Kaminsky & Dewey, 2001). Where one sibling has a disability, sibling relationships may be even more important to families.

Richardson and Jordan describe sibling relationships as ‘imperative to the lives of people with disabilities’ (2017: 1536). In a pragmatic sense, ‘positive’ sibling relationships may lead to typically developing siblings supporting and caring for their brother or sister with ID, taking over that role from aging parents. This is likely to become increasingly important in the UK context, with the decline of the welfare state and recent rising levels of poverty (JRF, 2017) along with improvements in the health and life expectancies in recent decades for people with certain IDs, such as Down syndrome (Hodapp & Urbano, 2007). There has been a systematic, thematic synthesis of qualitative studies exploring siblings’ lived experiences of having an autistic brother or sister (Leedham et al., 2020). The qualitative studies included in the synthesis provide further contextualisation about siblings’ relationships, experiences and outcomes. The synthesis also highlights that siblings across the studies spoke about the love, affection, and empathy they have for their autistic brothers and sisters. Understanding sibling relationships as an outcome in statistical modelling, and not just siblings’ psychological outcomes, is an important research focus that has been under-studied. Research that focuses on sibling relationships also disrupts the negative narrative about the effects that having a brother or sister with IDD has on the non-disabled sibling. Overall, there is little statistical evidence to support the assumption that sibling relationships where one of the siblings has an IDD are any worse than sibling relationships where neither sibling is disabled.

Summary of Empirical Literature

The existing empirical literature about siblings does include some population-level data. Where available, population-based data about siblings should be analysed as these data reduce issues related to sampling biases. The IDD sibling research field is still saturated with small-scale samples without normative samples with which to compare and contextualise findings. Survey questions should be carefully designed so that these comparisons can be made without the need to recruit ‘normative’ samples. Large-scale samples should be employed where possible to provide more power in analyses. Using larger samples provides further scope for more complex analyses, such as controlling for multiple predictors, and employing certain analysis techniques, such as structural equation modelling. As explored above, even when using high-quality population-based samples, findings about the outcomes of siblings were relatively mixed. This indicates that there may be a lot of variance in the outcomes of siblings. Studies about siblings tend to focus on direct effects, rather than on more complex analysis techniques. For example, exploring moderating effects may help us better understand under what conditions some siblings may experience better or worse outcomes than other siblings. The empirical data suggests that the importance of SES in understanding the outcomes of siblings is an important factor, this should be measured in a more developed way than merely income, as this can be contingent on geographical locale as well as household factors.

3. Reflexivity and ‘Insider’ Research

My brother is autistic and that experience has undoubtedly shaped my understanding of, and my initial interest in, this research. I have known since the start of my PhD that I would need to write something self-reflexive for this thesis, and although I have attempted it from time to time, I always ended up writing something vague and structural, distancing myself from the writing. I understand my reticence and reluctance. After all, this is not just my story to tell. In truth, it feels like it is barely my story to tell at all. Fundamentally, it is his. This is a difficult truth about family disability research – the very premise of our interest in the family as a whole, or siblings in the case of my research, is tied up in the identity of another person.

Reflexivity describes a process whereby researchers reflect on how their assumptions, biases, approaches and presence will affect their research, calling for us to interrogate hierarchies and power imbalances within our research (Potter & Hepburn, 2012). Self-reflexivity is defined as a process whereby we as researchers ‘critically interrogate ourselves and one another regarding the ways in which research efforts are shaped and staged around the binaries, contradictions, and paradoxes that form our own lives’ (Lincoln & Guba, 2003: 283). Therefore, it is an important process for all researchers, although it is more often explicitly explored by people with personal experience of the subject matter, and qualitative researchers. It can be used to validate, enhance understanding, and consider ethical issues in our research (Finlay, 2012). Being a sibling myself, I can be seen as an ‘insider’ (Hodkinson, 2006), as I share some experiences and characteristics with the participants of my research. This does afford certain benefits, such as being able to design and analyse research from another perspective and it may help to build rapport with participants. This identity does also carry risks related to allowing my own experiences and ‘un-elucidated prejudices [to] dominate’ (Finlay, 2003: 108) the research process. Therefore, self-reflexivity is a fundamental part of social research, particularly for those that have some sort of ‘insider’ status. As emphasised by Griffiths (1998), we are only ever partial, relative insiders. Although my experiences may provide me with some insights I would not otherwise have, it may also mean I risk making assumptions about others siblings’ experiences based on my own. This section will describe some of my reflections about my family and my relationship with my brother. I will then explore some of the issues and assumptions that are highlighted from my reflections that I feel are relevant to my research.

Childhood

I was seven when my brother was born, and he was diagnosed with autism relatively late, when he was 11. There were six of us at home, my mother and step father, myself, two sisters, and my brother. My brother as a child was, and still can be, intense – and I mean that in the best possible way. He would talk unceasingly, he would both demand and, rather skilfully, command whatever space he was in. He would get frustrated and volatile if things did not go his way – and would have meltdowns frequently. Our family would tip-toe around him to limit these outbursts. As for me, I was a nauseatingly well-behaved child, always trying to be helpful to

my parents. I could also be moody and sulky, and I talked too much. I write this not to be overly-solipsistic – but in fairness. If I am going to over-share traits about my brother, I ought to do so about myself as well. I was an anxious child anyway – but I remember fretting about my brother a lot when he was small. My brother had several seizures when he was a toddler that led to hospital stays. He would not eat enough and was much too thin. What he did eat was very restricted. When he started school, he would only eat his packed lunch if I made it for him in a very particular way. I was quietly thrilled to be helpful.

I would often feel quite embarrassed about my brother when I was a child. I remember having to help him with toileting – and my friends not understanding why I was being called in to help him. My brother did not look disabled and did not have a diagnosis – it was hard to explain to my friends why he needed extra help, because I did not know why he needed it myself. During sixth form I used to pick my brother up from school quite regularly to help my mum. I think she was also avoiding having to talk to his teachers at this time so it served a dual benefit. On some days he would have melt downs during the walk home. I had no idea how to help him or keep him safe – it was frustrating and exhausting, and I would dread running into a classmate. I would eventually get him back to our flat, and I do not remember ever feeling the need to tell my parents what had happened. It was so common and none of us really knew how to handle meltdowns at this time, so it felt like there was no benefit in discussing it. I feel ashamed to think I ever felt embarrassed about my brother – and I felt this shame both at the time of these incidents as well as whilst writing this. I think this shame has implications for whether and how siblings share their experiences.

Education

We moved every two years or so – and I went to ten schools in total. Sometimes, particularly before my brother started school, these moves were for more financial reasons. Later though, it was hard not to see that uprooting us all was my mother's response to the way that my brother was treated. My brother faced a lot of exclusion and unkindness, particularly as he got older, both at school, and from our wider family. My mother and step father faced a great deal of judgement too. Every move felt the same, my mother would feel hopeful and excited about a new start, and

then my brother's teachers would start to make comments or talk about how they were disciplining my brother. Other parents would make comments in the playground about him, and as my brother got older, the other children did not want to play with him. My mum would get very upset and withdraw. Looking back now I suspect she was intermittently depressed for a lot of my childhood. My step dad would keep things going at home and at work – sometimes literally covering my mum's shifts. Eventually another one of us would have a bad few weeks at school, and my mum would announce that we were moving again. These moves were very disruptive and drawn out. As we knew we were moving in the coming months, we would all withdraw further from school – sometimes literally by being 'home educated'.

For one move, I was sent ahead to live with an aunt to get started at a new school sooner, for another move, my older sister was left behind to live with my grandmother to finish an academic year. All of our attendance records were terrible – our mum let us stay home whenever we wanted, and I can remember her persuading me to stay home with her a few times too. I think she wanted the extra help and company, particularly when my brother was off. It may also have been due to my mother already having enough to deal with, and not wanting to fight with us about attending school. I think we all developed this habit from mum of retreating from and avoiding the world – and school became something that we felt was optional. It would be unfair to say the moves were always driven by my mum's desire to provide a better space for my brother, as I also remember a family move at about the time that he was born that seemed to be most driven by my anxiety around my new school.

Diagnosis and Support

I had just begun university when my brother was diagnosed. He was being home-schooled at this time because his teacher had physically hit him across the back of his head, apparently because he was not making sufficient progress on the task that he had been set. My family and I had never heard of the 'autism spectrum' before my brother was diagnosed and I am not sure if that was because we lacked the sort of capital needed for that awareness, or whether it was just that it was a lot less commonly known then. I expect it was a combination of both. The way that he was

treated seemed worse pre-diagnosis. Post-diagnosis, my mum and step-dad had the vocabulary in the form of a diagnostic label to push back against outsiders. I think my mother felt validated by the diagnosis and relieved that there would be extra support for him, although accessing extra support has been a constant battle.

Our Relationship

I think my relationship with my brother was, and is, in many ways similar to other older sisters with younger brothers. I remember in childhood having more conflict with my brother than my two sisters did with him and it would be dispersed with periods of mutual adoration – which I think is quite typical of sibling relationships as well. If I threw stones at his bedroom window at 2am after my waitressing shift, he would come downstairs to unlock the deadbolt on the door so I could come in without waking mum. I think what was different about our relationship was that it was not allowed to play out as organically without interference – as we were expected to all work together to make things as easy as possible for my brother. I can understand this well now, he had a very difficult time at school and needed support and compassion at home. It did mean however, that I often felt like his carer rather than his sister. Possibly that is completely ordinary given the age gap between us. I have mixed feelings though about this interference because I do think this has led to me detaching at times and avoiding conflict in an unhealthy way. In adulthood, a lot of my interactions with him make me feel overwhelmed. Our relationship is becoming increasingly reciprocal. The first time he asked me a question about me was only about five or six years ago. When our family gathers, I often catch him in the corner of my eye building himself up to join the family and interact with us – he tries so hard – and I wish I could make it easier for him. During COVID-19 he has independently called me during lockdown several times and even advised me on how to make progress on my thesis, (take lots of breaks apparently) and told me that he was proud of me for trying whether I finished it or not. I can see how reciprocal our relationship has become and how important our relationship is to both of us, even if it may not seem that way to other people.

Ethically, it is challenging to talk about someone that has not consented to be spoken about. I have been able to speak with my brother a couple of times about my PhD and about me talking about him. This is not a conversation that all siblings of

people with IDD are able to have. Recently, I spoke with him about writing this section and dedicating my thesis to him. I also asked him how he felt having neuro-typical sisters. He told me that he finds it hard to understand what we mean or what we are thinking, which is something I have heard siblings say about their brothers and sisters with IDD. He explained that he liked having three sisters because he always had someone to talk to – as he often did not have friends at school. I think a lot of sibling research focuses on siblings being a source of care and support, but my experiences make me feel, or perhaps assume, that one of the most fundamental roles siblings can fulfil for one another is that of a friend.

Poverty and Structural Factors

Things were harder for my family than they ought to have been. We grew up poor. My family lived very much hand-to-mouth, and we did not always have enough to eat. Our gas and electricity meter would often run out of funds and we would rummage the settee hoping to find enough shrapnel to put it back on. I remember once when I was about nine or 10, my mother asking me whether she should buy us some food for dinner or put the heating and electricity back on. We chose dinner. My mother set us up a carpet picnic in the lounge, with candles for light and all our duvets around us for warmth, and we made it a game for my younger siblings. It would be impossible for me to even begin to explain our childhood without disclosing that we were living in poverty for much of it. Being as poor as we were was incredibly stressful for me, and I know the stress I felt was nothing compared to the stress my mother and step father would have felt about it. If I think about my childhood and what made it hard, it was poverty first and foremost, it was never because of my brother. I can now reflect however on how there might have been some indirect factors related to having an autistic brother. My mother never worked more than part-time hours for example, and this may have been related to my brother's needs. I also wonder whether class biases played some role in the reason my brother was diagnosed quite late. Population-level data also tell us that families of people with ID are more likely to experience poverty and low socio-economic status (Emerson, 2003; Emerson, 2004), therefore my experiences are more common in families like mine. I can also appreciate that I grew up predominately under a New Labour government, and so it concerns me that the

poverty I experienced is both worse and more common now than it was when I was growing up.

I have mentioned the constant battle my parents faced, and continue to face, to help my brother get the support that he is entitled to. My mother and step father both left school at 16 and so there was a steep learning curve for them to acquire the skills required to navigate service provision. In my twenties, my mother would invite me to meetings with my brother's college when she was worried that they wanted to avoid providing him with the support that he needed to progress onto the next stage of his course. She felt that his course tutors would take me more seriously than her, due to the assumptions people make about her social class and mine. Navigating service provision for disabled children and adults is a challenge for all families, but I expect that lower-levels of education combined with the judgements that often middle class professionals make about working class families can make this process harder for some families.

It is hard for me not to see my brother's and our family's experiences outside of a social disability model and a more structural framework. Most of his problems, and my family's, stem from my brother's interactions with the outside world or structural inequalities. There were things that were difficult about my childhood, and it would be oversimplifying them to say that they were because of my brother. My mum was stressed and I think probably depressed, but if she had the right support from services and communities outside of our household, then things would have been much easier. I think this led my mum to lean on me and my sisters a lot more than she ought to have done. A lot of my more negative experiences about being a sibling to someone with autism in my view, stem from living in poverty for much of my childhood. Most of my fears for my brother's future stem from my lack of confidence that we will have the economic resources we need to support him to have a good quality of life in the future.

Methodological Issues Raised from Reflections

Self-reflexive practices are seen as essential in qualitative research, yet I have not seen it mentioned as a practice for quantitative social science research. I approach all social science research from an interpretivist and social constructivist perspective. My own experiences and assumptions will affect how I design my

research, how I collect data, how I analyse data, and how I interpret my findings. There are countless decisions I have made throughout my research and these have been guided by my own experiences. Therefore, self-reflexive practices have an important role in quantitative research. This section has highlighted some assumptions I have based on my personal experiences, some of which are well founded in the empirical literature, such as the effect of poverty and socio-economic deprivation on families of people with IDD. I also assume that having multiple typically developing siblings would be a protective family characteristic, although the empirical basis for this is very mixed. I doubt whether we can seek authentic and truthful data in social research. I have mentioned my concern with, for example, my mother not agreeing with some of my interpretations of my childhood. It is part of the reason I think having multiple siblings is a benefit, because I have peers that I can discuss our childhood with and we can make sense of it together. Self-reflexive qualitative approaches help us to understand ‘that all lives are composed – that the stories of our lives are indeed constructed, fabricated, invented, made up...’ (Plummer, 2001: 400). This, according to Plummer (2001) does not make self-reflexive accounts and narratives deceptive or methodologically invalid, but reveals its value, with participants’ interpretations leading to ‘different kinds of “truths”’ (Plummer, 2001: 401). Thinking about self-reflexive accounts in this way is helpful both in settling some of my concerns about writing this section, and also for reflecting on the sort of data we receive from our participants.

Self-reflexivity has been considered culturally biased, unscientific, and un-objective (Reed-Danahay, 2001). Approaching quantitative research with a more interpretivist framework helps us understand that all research has these problems, qualitative research is just more honest and open to this fact. The difficulties I had writing this section, and how heavily edited and processed it is, has implications for the data we receive from our participants. The temptation when sharing personal experiences is to err on the side of positivity, and this is something I have noticed when reading or hearing other siblings’ reflections about being a sibling or a family member of a person with IDD. I understand the reason for this reticence – I think siblings have a desire to disrupt the negative narrative associated with having a brother or sister with an IDD. I have also noticed this reluctance to talking publicly about sibling experiences with other siblings I know. There are things we say to one

another, and then there are things we say to non-siblings. This is relevant, because it raises important epistemological issues in our research. What is sayable and unsayable within these research exchanges? What will our participants share with us, and what are they not telling us? What are the implications of these gaps in our knowledge? This is an issue in all social research, but within IDD family research the implications are particularly complex, because we are including people in our research, by merit of their relationship with another person. The problematic nature of our work is exacerbated by our social context, so often emulated in our research, which excludes and marginalizes people with IDD.

There are further ethical issues related to insider research, such as being unable to anonymize myself, and by extension, my family. As highlighted, there are ethical issues related to using insider-status as an ‘authority move’ (Chapman Houlton, 2012) and by coaxing participants to share more than they otherwise would (Griffiths, 1998). There are questions about self-disclosure that have been discussed by therapists (Audet & Everall, 2010; Rogers, 1961; Audet, 2011), whereby self-disclosure is only seen as appropriate if it advances and helps the therapy process. Working with families and siblings throughout my PhD through participant engagement, such as by collaborating with colleagues at UCL on the Sibling Research Advisory Group, or by working with Sibs, has shown me that sharing my sibling-status has not provided me complete ‘insider status’. My motives have still been questioned explicitly by family members, such as when I was asked by a 12 year old sibling whether I was trying to prove that other siblings felt the same way as I did. I can recall meeting three siblings where the eldest had autism and ID and the two younger siblings were primary-aged children without disabilities. The first time I met with them, the youngest boy kept bursting out to say another good thing about having a brother with autism and ID. When I met them a second time, and spoke with the younger sister one-to-one, she told me that her younger brother had coaxed her to only say positive things, and that if she had to say something negative to me, to just tell me that their brother with autism and ID was a bad singer. For me, this reflects the way in which siblings, from an incredibly young age, seek to disrupt a negative narrative about IDD. In this example, one sibling felt social pressure from another sibling not to speak openly. It indicates to me that even though I had disclosed my sibling status, I was deemed an outsider. The family had discussed

together the narrative that they would tell me. Just as I have in writing this piece of prose, they edited and constructed a narrative that they were comfortable sharing with me. This has obvious implications for qualitative research, but it also has implications for quantitative research, as some participants may hold back on revealing certain aspects of themselves and their experiences when participating in, for example, surveys. Furthermore, these issues may result in some siblings not taking part in sibling research at all.

My final point about this personal reflection is that I can see how my own experiences fit into and can be contextualized by the theoretical perspectives already described. Throughout this chapter I have returned regularly to the need to disrupt a negative narrative, about the perceived negative effect that having a family member with IDD can have on the family system. However, things were hard for our family, and so I also believe there is still value in asking questions about siblings' psychological outcomes and examining why some siblings are struggling and some are doing well. I understand that our experiences as a family were and are more complicated than assuming that things were harder for the rest of our family because my brother was autistic. There were and are multiple, interacting, reciprocal factors that could explain our experiences as a family and as individuals. We need to ask better questions and apply more nuanced statistical techniques to examine under what circumstances some siblings fare better than other siblings.

Thesis Structure

The broad structure of this thesis includes four empirical chapters followed by a general discussion. These four empirical studies have been written in the form of distinct, academic journal articles. Therefore, the composition of each empirical chapter is written independently, including an orientation of the existing literature relevant to the chapter, followed by methods, results and discussion sections within each chapter. Two of these studies have already been published in peer-reviewed journals (Hayden et al., 2019a; Hayden et al., 2019b) and two of them are currently being prepared for submission to journals. Combined, these studies contribute to our understanding of the outcomes of both child and adult siblings of people with disabilities. Chapters two and four consider psychological outcomes for child and adult siblings and chapter three evaluates a potential source of support for child

siblings. Chapter five begins to move beyond psychological outcomes and consider sibling relationships.

Chapter two draws on population-level data from the Millennium Cohort Study to explore the behavioural and emotional outcomes of older siblings of children with ID ($n = 257$) compared to older siblings of children without ID ($n = 7246$). We aimed to understand using representative data whether siblings were more likely to experience worse outcomes, and if so, would these group differences remain once we controlled for other predictors. This study provided the first population-representative data about the outcomes of siblings of children with ID.

Chapter three considers potential supports for siblings of children with disabilities, discussing school-based factors for siblings. Embedded in this chapter is an evaluation of a school-based intervention designed by Sibs for primary school children that have a brother or sister with a disability ($N = 55$). This was my only study that was not specifically about siblings of people with IDD. However, the majority of the sample did have a brother or sister with an IDD. What was unique about this intervention is that it was school-based and one-to-one between the sibling and a staff member from their school.

Chapter four provides findings from the Adult Sibling Survey, a large-scale survey of 911 adult siblings of people with IDD. The study was co-designed with Sibs and we explored the mental distress, wellbeing, quality of life, and health outcomes of adult siblings. As we had used validated measures that had been used in existing national surveys, we were able to first compare these data to population 'norms' – something that has rarely been done in adult sibling studies. We then used moderated regression analyses to consider whether the association between sibling carer status and the sibling outcomes were moderated by socio-economic position and/or the level of independence of their brother or sister with IDD. This analysis technique allowed us to move beyond direct effects and consider under what conditions siblings experience different outcomes.

Chapter five presents sibling data from the 1000 Families study (Hastings et al., 2020). Again, the data from this study were large-scale, with over 1,000 Families recruited in the overall study sample. Once we identified those cohort children with a sibling that did not also have an IDD, we had a sample of 500 sibling dyads: with

one sibling with ID, and one sibling without an IDD. This study moves beyond more psychological outcomes of siblings and explores sibling relationships, an important area of research that is relatively under-explored in the existing literature in comparison to research about siblings' psychological outcomes. The analysis and conceptualisation of this study were relatively unique. We used structural equation modelling to explore relationship outcomes of the siblings by considering the behavioural and emotional symptoms of both siblings. Existing research tends to focus on these behavioural and emotional symptoms as outcomes, as we have in prior chapters. By including the behavioural and emotional symptoms of both siblings, we have explored reciprocal effects and attempted to disrupt the narrative that it is the child with ID that is negatively impacting the sibling without IDD.

Chapter six provides an overall discussion synthesising the main contributions of these four empirical chapters. I revisit some of the issues and ideas introduced thus far, and provide a critical reflection of both my work, as well as wider issues within the IDD family field.

Chapter 2: Study 1 - A population-based study of the behavioural and emotional adjustment of older siblings of children with and without Intellectual Disability¹

¹ This study has been published in the *Journal of Abnormal Child Psychology* as follows: Hayden, N. K., Hastings, R. P., Totsika, V. & Langley, E. (2019a) A population-based study of the behavioral and emotional adjustment of older siblings of children with and without Intellectual Disability. *Journal of Abnormal Child Psychology*. 47, 8. 1409-1419. doi: 10.1007/s10802-018-00510-5. The Centre for Longitudinal Studies, UCL Institute of Education, the UK Data Archive and UK Data Service make MCS data available for secondary analysis. However, they bear no responsibility for the analysis or interpretation of these data.

Chapter Two: Study One

The question of whether siblings of children with disabilities have worse psychological outcomes compared to siblings of children without a disability has been relatively well-explored in the existing ID research. However, these studies have often employed small-scale convenience-based samples that are more biased and, therefore, inconclusive. Population-based data overcome some of these issues, being large-scale and representative. Although there are existing studies about the outcomes of siblings of children with disabilities more generally (Goudie et al., 2013; Neely-Barnes & Graff, 2011; Emerson & Giallo, 2014), this chapter provided the first published population-level data about siblings of children with ID specifically (Hayden et al., 2019a). Since publishing this study, Canadian based population-level data have also emerged about siblings of children with DD (Marquis et al., 2019; Marquis et al., 2020). These studies have been discussed in chapter one and I will revisit their work in my overall discussion (chapter six). Our study presented in this chapter benefits from having access to a wide selection of covariates to include in our analyses. For example, we were able to include a range of measures about the families' SES, and we were able to include family-systems level variables, such as about the behavioural and emotional outcomes of their brother or sister with ID, the mental distress of the mother, and whether they were living in a single parent household. Rather than mental health diagnoses, as in Marquis' work, our work was about siblings' behavioural and emotional problems and prosocial behaviour, measured using the Strengths and Difficulties Questionnaire (Goodman, 1997). This allowed us to understand both the average, as well as clinical cut-offs, for problem behaviours and emotional well-being. This measure has been used consistently in this thesis, in each of the child sibling empirical studies (Chapter 2, 3, and 5).

Abstract

This is the first study on the behavioural and emotional adjustment of siblings of children with intellectual disabilities (ID) to use a population-based sample, from the third wave of the Millennium Cohort Study (MCS); a UK longitudinal birth cohort study. We examined differences between nearest-in-age older siblings (age 5-15) of MCS children (likely mainly with mild to moderate ID) identified with ID ($n = 257$ siblings) or not ($n = 7,246$ siblings). The Strengths and Difficulties Questionnaire (SDQ) measured all children's adjustment. For SDQ total problems, 13.9% of siblings of children with ID and 8.9% of siblings of children without had elevated scores (OR 1.65; 95% CI 1.04, 2.62; $p = .031$). Similar group differences were found for SDQ peer and conduct problems. In logistic regression models, variables consistently associated with older sibling adjustment were: adjustment of the MCS cohort child, older sibling being male, family socio-economic position, primary carer psychological distress, and being from a single parent household. The ID grouping variable was no longer associated with adjustment for all SDQ domains, except siblings of children with ID were less likely to be identified as hyperactive (OR 0.30; 95% CI 0.10, 0.87; $p = .027$). Some older siblings of children with ID may be at additional risk for behavioural and emotional problems. Group differences were related mainly to social and family contextual factors. Future longitudinal research should address developmental pathways by which children with ID may affect sibling adjustment.

Introduction

Although there is a range of existing research exploring developmental outcomes for children who have a brother or sister with a disability, including intellectual disability (ID), current research is both inconsistent and contradictory in answering whether these siblings of children with ID are at an increased risk of poorer outcomes – as might be predicted from family systems theory (Kovshoff et al., 2017). For example, an early meta-analysis found that siblings of children with ID had more psychological and social problems than comparison groups, although these group differences were small (Rossiter & Sharpe, 2001). In more recent studies, researchers have reported similar relatively small group differences but also considerable variability in outcomes. A number of studies have identified more behavioural and emotional problems in siblings of children with disability compared to other children or to normative samples (Hastings, 2003; Verté et al., 2003; Ross & Cuskelly, 2006; Orsmond & Seltzer, 2007; Goudie et al., 2013). Other researchers found little or no group difference (Cuskelly & Gunn, 2006; Hastings, 2007; Howlin et al., 2015).

There is a distinct lack of representative population-based studies in this area. The strength of such studies is they are less affected by referral or self-referral biases and may allow conclusions about the whole population of siblings, or the whole population of siblings of children with ID. We found three population based studies of siblings of children with disability, but no population-based studies focusing on siblings of children with ID specifically. Focusing on ID specifically is important since different disability profiles or diagnoses have been shown to be associated with varying impact on the family system including siblings and parents (Hastings, 2016). In addition, key putative risk factors for sibling outcomes such as the behavioural and emotional adjustment of the disabled child and parental psychological distress are more prevalent in families of children with ID compared to other families of children with other disabilities (Hastings, 2016).

Goudie et al. (2013), analysing data from the USA Medical Expenditure Panel Survey, identified 245 siblings of children with disability and 6,564 siblings of children without disability. Disability in this study included those with cognitive disabilities, physical disabilities and chronic conditions. Goudie et al. (2013) found

siblings of children with disability had more social problems, problems with behaviour, and problems in school. For example, siblings of children with disability were 2.77 times more likely to have significant levels of problem behaviour.

Emerson and Giallo (2014), using a nationally representative group of children in Australia, explored the psychological well-being and adjustment of siblings of children also with a broad range of disabilities. There were 7,636 children included in the analysis who were living with at least one sibling for both study waves included in the research that did not have a brother or sister with a known disability, and 286 children had a brother or sister with disability. Emerson and Giallo (2014) initially found that siblings of children with disability had lower well-being than the group of siblings without a disabled brother or sister in some, but not all adjustment areas. However, once analyses controlled for the effects of socio-economic deprivation and other associated hardships, the small group differences in well-being were no longer statistically significant. Thus, the putative impact of child disability on sibling outcomes may have been mediated via socio-economic deprivation or explained directly by exposure to poverty. Such competing hypotheses could not be tested in the context of the cross-sectional methods used.

Neely-Barnes and Graff's (2011) study in the USA using national health data showed similar results when measuring psychological outcomes of siblings of children with disabilities. Neely-Barnes and Graff (2011) identified 373 siblings of a child with a disability and 3,790 eligible siblings of children without a disability. Between-group differences were non-significant once 12 additional factors were controlled. Again, co-occurring risk factors such as low income appeared to explain the association between child disability and sibling outcomes. However, causal effects could not be established within this cross-sectional design.

The Goudie et al. (2013), Emerson and Giallo (2014) and Neely-Barnes and Graff (2011) studies explored disabilities defined broadly and did not focus specifically on ID. The latter two studies do indicate the importance of exploring alternative factors that may affect sibling outcomes either directly or that may be associated with having a disabled brother or sister. In particular, socio-economic deprivation and economic factors affecting families are key variables. Emerson and Giallo (2014) also included maternal mental health in the variables they examined

(as would likely be important from a family systems perspective), but the other population-based studies did not. Thus, there is still no population-based analysis of siblings of children with ID from a dataset that also allows the exploration of the impact of a number of correlates of sibling outcomes and examined for their independent effects. Furthermore, the three existing population based studies differ in their findings. Therefore, further work is needed not only with an ID focus, but to understand more about correlates of sibling behavioural and emotional adjustment.

Factors other than socio-economic variables might also affect siblings' behavioural and emotional adjustment. These include: the age and sex of the sibling (Hastings, 2003; Verté et al., 2003; Cuskelly & Gunn, 2006, Orsmond et al., 2009; Petalas et al., 2009; Walton, 2016); the sex composition of sibling dyads (Cuskelly & Gunn, 2006); birth interval (Martin & Horriat, 2012); the number of brothers and sisters in the household (Burke et al., 2012; Goudie et al., 2013; Walton, 2016), and whether the family is a single parent/carer household (Deater-Deckard & Dunn 2002; Kelly et al., 2009; McHale et al., 2012).

More importantly, in addition to socio-demographic factors, a family systems perspective on outcomes for siblings of children with disability (Kovshoff et al., 2017) suggests that the well-being of other family members is likely to affect sibling psychological adjustment. Consistent with this systems perspective, a number of studies have shown associations between maternal psychological distress and sibling adjustment (Quintero & McIntyre, 2010, Petalas et al., 2012), and between the behavioural and emotional problems of children with ID and their sibling's psychological adjustment (Hastings, 2007). Existing population-based studies of siblings of disabled children have not examined the independent associations of additional putative risk factors such as parental psychological distress and the siblings' brother or sister's behavioural and emotional adjustment in addition to a range of socio-economic and demographic factors.

The aims of the present study were, therefore, to: (i) explore if there were group differences in behavioural and emotional adjustment for siblings of children with and without ID in a nationally representative sample, and (ii) to explore, if there were differences, which correlates identified from the existing literature were associated with sibling adjustment. These two research questions extend existing

literature through the focus specifically on ID, in a population-representative sample, and incorporating a wider range of known correlates of sibling adjustment.

Method

This study is a secondary analysis of data from the third wave of the UK's ongoing Millennium Cohort Study (MCS3; 'Millennium Cohort Study: Third Survey', 2017). MCS is a longitudinal birth cohort study following the lives of 19,000 children born in 2000-2001 (MCS, 2017), with MCS3 following up with cohort member children at age five years. Cohort children are identified through Child Benefit Records – a non-means tested state universal benefit with a very high uptake among UK families with children at the time of the MCS inception. Cohort member children were randomly selected from those children eligible to receive Child Benefit, living across the UK, at age nine months, and born between September 2000 and August 2001 (Plewis, 2007). A two-stage stratification was followed to ensure that a nationally representative sample with adequate representation from ethnic minority and disadvantaged children was achieved. Weights were subsequently developed to account for the MCS sample design (Hansen, 2012). In the present study, to ensure the sample is a nationally representative sample, data were analysed with weightings through complex samples procedures (Jones & Ketende, 2010).

In the present study, the data analysed were collected from the primary respondent. This was the mother (biological or adoptive) in 99.8% (weighted) of families. The remaining primary respondents were also included in the analysis and included fathers (natural, adoptive or step) as well as other extended family members. Primary respondents will be referred to as primary carers throughout this paper. Data about the MCS cohort members' older siblings (but not younger siblings, given the young age of the cohort children) were collected in both waves two and three of the MCS, although the focus of this paper is on data from the latter study (MCS3) at which time index child and their siblings will have had five years of life together.

We explored group differences between the nearest-in-age older siblings (aged five to 15 years) of those MCS cohort member children identified as not having (n of siblings = 7,246) or having (n of siblings = 257) ID. Those siblings of

children who could not be classified as having or not having ID were not included in the analysis. For this study, cohort children with ID were identified using a grouping variable adopted in previous research (Totsika et al., 2018). To identify cohort children with ID, data from the second, third and fourth waves of the MCS were used through a four-step process. At age seven, trained interviewers assessed children's word reading and pattern construction skills, two scales from the British Ability Scales (BAS-II; Elliott et al., 1996) along with mathematics ability (NFER Progress in Maths). A factor analysis of the age standardized scores of these measures provided a total cognitive ability index g that accounted for 63% of the total variance across these measures. Intellectual disability was defined as a score two standard deviations below the mean of the total cognitive ability index. A total of 352 children were classified as having ID (g scores <70) using these age seven variables. Age seven was selected as the age of first choice to identify ID because cohort children would be around two years into their formal education in the UK and at an age when identifying children with ID is arguably ideal (Maulik et al., 2011). For those children unclassified at age seven (e.g., because test data were missing or their family did not respond to MCS at wave four), cognitive test data (BAS-II; Elliott et al., 1996) at age five were used in a similar way. This second step identified a further 137 children with ID. For children that remained unclassified following the first two steps, age-standardized scores on the Bracken School Readiness Assessment –Revised (Bracken, 1998) at age three were used. A fourth step used parent and teacher reported information at age seven. Where both parent and teacher had independently indicated that the cohort child had special education needs, and additionally the teacher reported that the cohort child was performing significantly below average on five academic outcomes associated with reading, writing and maths, then the child was classified as having an ID. A further 17 children were identified as having ID at this step.

The four step classification process resulted in 555 cohort children being identified as having ID from a total sample of 19,244 MCS children (equivalent to 2.7% weighted, or 2.9% unweighted, of the MCS sample). Of these MCS children identified as having ID, 257 had one older sibling aged 5-15 at wave three with suitable data available for analysis. Although information was provided on more than

one older sibling where applicable, we only included one available older sibling, nearest in age to the cohort member, in the analysis.

Participants

Demographic characteristics of the identified older siblings of children with ID and children without ID are summarized in Table 2.1. There are no statistically significant group differences in these characteristics for older siblings of children with and without ID for the following measures: the older sibling being male; older sibling and cohort member being the same sex; the age of the cohort member; and the age difference between the older sibling and the main cohort member. Siblings of children with ID were more likely to be living in a single parent household ($\chi^2(2, N = 7,783) = 27.80, p < .001$), be from a family who experienced more socio-economic deprivation ($\chi^2(1, N = 7,763) = 102.08, p < .001$), and have a primary parent or carer experiencing psychological distress ($\chi^2(1, N = 7,458) = 6.27, p = .013$). In addition, older siblings of children with ID were more likely to have a younger brother or sister (the main cohort member) with elevated SDQ (Strengths and Difficulties Questionnaire; Goodman, 1997) total difficulties scores ($\chi^2(1, N = 5,186) = 127.57, p < .001$). The number of children in the household was higher for households of a child with ID ($t(1, 389) = -5.88, p < .001$). In addition, siblings of children with ID were more likely to be older ($t(1, 389) = -2.90, p = .004$).

The primary respondent was usually the natural parent of the cohort member child (for 99.6% of cohort members) and was typically the mother (97.9% of primary respondents were female) for both those families with and without a child with ID. Primary respondents of a cohort member with ID were younger (mean age = 33.39; $SD = 6.19$; Range = 26 compared to the primary respondents of a cohort member without ID (mean age = 35.21; $SD = 9.57$; Range = 42 ($p = .002$). It was not possible to determine if the older siblings themselves had an intellectual disability from the data available from the MCS for this secondary data analysis. There was also no genetic information available for siblings, including no report of autism or other diagnoses for siblings.

Measures

Behavioural and Emotional Adjustment.

The behavioural and emotional adjustment of siblings was measured using the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997). Main respondents were asked to complete the SDQ about the older sibling. The SDQ includes 25 items to assess the psychological adjustment of young people and children (Goodman, 1999) using a three-point rating scale (i.e. ‘not true’, ‘somewhat true’ or ‘certainly true’) to assess the extent to which the statement applies to the child. Items include statements such as the older sibling being ‘considerate of other people’s feelings’ and ‘often unhappy, down-hearted or tearful’. The items represent five distinct scales: emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behaviour. A SDQ total difficulties score is derived by summing the first four sub-scores (excluding prosocial behaviour). For the purposes of this study, SDQ scores were also dichotomized at the advised clinical cut off levels (Goodman, 1999) for ‘normal’ and ‘abnormal’ scores. Primary respondents also completed the SDQ for the cohort child (the MCS cohort child with or without ID).

Primary Carer Psychological Distress.

To measure the psychological distress of the primary respondent, usually the biological mother, MCS used the K6 self-completion measure (Kessler et al., 2002). The K6 asks the person completing it how often they have felt in the last 30 days ‘nervous’, ‘hopeless’, ‘restless’, ‘fidgety’, ‘so depressed that nothing could cheer you up’ and whether everything was an ‘effort and worthless’. Responses are based on a five point rating scale to measure the extent to which each question applies to the respondent. These items are summed to derive a 0 to 24 score. For the purposes of the present study, this scale was dichotomized with primary carers scoring 13 and above (which has been identified as a reliable cut-off for identifying serious mental illness) versus below a score of 13 (Furukawa, 2003; Kessler et al., 2010).

Socio-Economic Deprivation.

To measure the socio-economic deprivation experienced by MCS families at wave three, a composite variable was produced. This variable was based on previous research (Totsika et al., 2011; Totsika et al., 2013; Totsika et al., 2014; Totsika et al., 2015). Five indicators were incorporated to form this composite measure. Subjective poverty was measured on a five point rating scale identifying how well families felt

they were managing financially. Responses from primary respondents were dichotomized into families finding it ‘quite’ or ‘very difficult’ to manage financially versus families who were managing financially. MCS3 included five items about material deprivation by enquiring if families had access to basic material goods relevant in the UK, such as a weatherproof coat for the cohort member, two pairs of all-weather shoes, or if they were able to afford an annual holiday without staying with relatives. Older siblings’ families were grouped into those who could not afford two or more of those items versus those families that could afford all or all but one of those items. The economic activity of families was dichotomized by workless families versus families with at least one parent or carer working. Income poverty was measured using the OECD’s definition: families with an income below 60% of UK median equivalized income versus families with an income above this level. Neighbourhood deprivation was measured using the UK Indices of Multiple Deprivation (IMD) information which measure deprivation for small geographical areas based on seven different domains of deprivation derived from national Census data. This includes measures of income, employment, education, health, crime, housing, and environment (Gill, 2015). Neighbourhood deprivation rankings for England, Northern Ireland, Scotland and Wales were incorporated into one variable and dichotomized by whether families lived in a neighbourhood in the bottom (most deprived neighbourhoods) quintile versus all other quintiles for their UK country.

The five dichotomized indicators were summed into one socio-economic deprivation composite measure. The composite measure values ranged from zero to five with higher values relating to higher levels of deprivation for families. This summed scale was then further dichotomized to identify those families experiencing socio-economic deprivation (those with one or more indicators of socio-economic deprivation) versus those families experiencing no indicators of socio-economic deprivation. This dichotomization was used because almost 50% of families had no indicators of socio-economic deprivation, and so the summed score had a highly skewed distribution that was otherwise difficult to transform.

Procedure and Analysis Approach

The data for this study were from MCS3, available from the Centre for Longitudinal Studies, UCL: IOE (‘Millennium Cohort Study: Third Survey’, 2017).

Data were available to download from the UK Data Service. Ethical approval for MCS1 was gained by the original investigators from the South-West Multi-Centre Research Ethics Committee and MCS2 and MCS3 was gained from the London Multi-Centre Research Ethics Committee. All adult respondents provided informed, written consent to take part in the MCS study for their own involvement and also as parent/guardian of the participating child/children. To download the data, researchers must register and agree to a number of data privacy conditions, including maintaining the confidentiality and anonymity of the families included in the study. In addition, for the present secondary data analysis, ethical approval was granted from the University of Warwick's Humanities and Social Sciences Research Ethics Committee as per institutional requirements (see Appendix One, 1.1-1.2 for institutional ethics documentation). Analysis was performed using SPSS version 24©. The sample weightings required to ensure the sample was representative of the UK population meant that all analysis was performed through SPSS complex samples options (Jones & Ketende, 2010; IBM Software Group, 2012).

Our first research objective was to explore whether there were group differences in the SDQ total difficulties score and SDQ sub-scale scores between those older siblings of cohort member children with ID and those older siblings of cohort children without ID. This was explored and analysed using t-tests through general linear models. Group differences in the proportion of siblings scoring in the 'abnormal' range on SDQ scores were also explored using Odds Ratios. By exploring this question in both ways we were able to not only explore general group differences, but also to consider the differences between those siblings with more concerning levels of behavioural and emotional problems (i.e. those scoring above the clinical cut off for scores on the SDQ).

We then explored whether any group differences would remain once socio-economic, demographic, and family factors were controlled and which of these factors were associated with siblings' behavioural and emotional adjustment. Using dichotomous scores for SDQ domains ('abnormal' range vs. not) as outcomes, logistic regression models were fitted to examine group differences alongside the following factors: sex of the older sibling, number of siblings and cohort member(s) in the household, age difference between cohort member and older sibling, the age of the older sibling, same or different sex for sibling pairs, family socio-economic

deprivation, single parent household, primary carer psychological distress (typically maternal psychological distress; K6), and the behavioural and emotional adjustment of the MCS cohort member child (SDQ total behaviour problem score). Logistic regression was selected rather than ANCOVA to explore the second research question as we were interested in correlates of older siblings' behavioural and emotional adjustment and specifically in those older siblings experiencing elevated SDQ scores who may represent the most important at-risk group of siblings from a clinical perspective.

Results

Sibling Behavioural and Emotional Adjustment Group Differences

Mean scores for primary carer SDQ ratings of siblings of children with ID and without ID are presented in Table 2.2. The SDQ total problems ($t(1, 389) = -2.97, p = .003, d = .19$), peer problems ($t(1, 389) = -3.85, p < .001, d = .26$) and conduct problems ($t(1, 389) = -3.46, p = .001, d = .22$) scores were higher for older siblings of children with ID. Cohen's d estimates indicate small to very small effect size differences between the groups. No statistically significant group differences were found for hyperactivity ($t(1, 389) = -1.65, p = .101, d = .09$), prosocial behaviour ($t(1, 389) = 1.48, p = .139, d = .09$), and emotional problems ($t(1, 389) = -0.68, p = .495, d = .05$).

Table 2.3 presents Odds Ratios for comparisons of older siblings of children with and without ID in terms of SDQ scores above the 'abnormal' range cut-offs. The siblings of children with ID were more likely to have elevated SDQ scores than the siblings of children without ID for total problems (OR = 1.65; 95% CI 1.04, 2.62; $p = .031$), peer problems (OR = 2.01; 95% CI 1.37, 2.95; $p < .001$), and conduct problems (OR = 1.75; 95% CI 1.19, 2.57; $p = .004$). No statistically significant group differences were found for elevated levels of hyperactivity (OR = 0.65; 95% CI 0.33, 1.30; $p = .217$), limitations in prosocial behaviour (OR = 1.23; 95% CI 0.85, 1.79; $p = .269$), and emotional problems (OR = 1.21; 95% CI 0.70-2.10; $p = .500$).

Logistic Regression Analyses

The results of the logistic regression models are summarized in Table 2.4. Logistic regression models were used to predict elevated ('abnormal' range) scores

for each of the five SDQ sub scores as well as the total difficulties score. For each of these six logistic regression models, socio-economic deprivation, elevated behaviour problems for the MCS cohort member child (i.e., whether the cohort child's SDQ total score was in the 'abnormal' clinical range), and the older sibling being male were statistically significant predictors of whether older siblings had elevated SDQ scores. Primary carer psychological distress, and being from a single parent household were also statistically significant predictors in most of the regression models.

After taking into account all other correlates, sibling group membership (siblings of children with/without ID) was not associated with older siblings' SDQ scores in five of the six regression models. The exception was for SDQ hyperactivity scores. The odds ratio for older siblings of children with ID having raised hyperactivity levels was 0.30 ($p = .027$; 95% CI 0.10, 0.87), indicating that the odds of them having increased hyperactivity levels was lower than for siblings of children without ID after accounting for other factors.

Discussion

The present study explored whether there were group differences in behavioural and emotional adjustment between older siblings of children with and without ID using a UK population-representative sample. In univariate analyses, statistically significant group mean differences were found for some, although not all, domains of the SDQ (total problems, peer problems, and conduct problems), where siblings of a child with ID had more problems than their peers. Effect sizes for these group differences were small. Using Odds Ratios to examine the proportion of each group meeting clinical cut-off scores on SDQ domains, older siblings of children with ID were approximately 1.5—2 times more likely to have problems in the 'abnormal' range on three SDQ domains (total problems, peer problems, and conduct problems) but did not differ on the other three SDQ domains. These results show that older siblings of children with ID have increased (small effect sizes) total problems, peer and conduct problems compared to older siblings who do not have a brother or sister with ID. Although some older siblings of children with ID have elevated SDQ scores compared to siblings of children without ID, differences in

clinical levels of behavioural and emotional adjustment problems relate to a small group of older siblings with particularly poor adjustment.

Once additional factors, informed by existing literature, were included in logistic regression models, the ID group variable was not significantly associated with sibling behavioural and emotional adjustment except for one SDQ domain (hyperactivity). These findings suggest that older siblings of children with ID are not at an increased risk of behavioural and emotional adjustment problems due simply and directly to having a brother or sister with ID. These findings are largely consistent with Emerson and Giallo's (2014) and Neely-Barnes and Graff's (2011) research exploring sibling outcomes in non-UK national samples and focused on mixed disability groups.

The demographic and family factors included in Table 2.1 indicate that older siblings with a brother or sister with ID were more likely to be from a family experiencing socio-economic deprivation, a single parent household, for their primary parent/carer to be experiencing psychological distress, and their MCS cohort brother/sister had elevated behavioural and emotional problems. These variables are risk factors for poorer well-being for children in general. In addition, older siblings with a brother/sister with ID were older themselves, lived in larger families, and had younger primary carers compared to other older siblings.

In the logistic regression models, being a male older sibling, coming from a family experiencing socio-economic deprivation, living in a single parent household, having a brother/sister with elevated behavioural and emotional problems, and having a primary carer with high levels of psychological distress were all consistently and independently associated with sibling emotional and behavioural adjustment. Therefore, an array of family and social factors in particular were associated with sibling adjustment (cf. Kovshoff et al., 2017). Accounting for these variables, and other demographic factors, reduced the initial sibling group differences to be outside of the range of statistical significance. The findings extend those from previous research beyond a focus on socio-economic factors to broader family systems issues and reinforce the importance of considering sibling adjustment from a multi-layered systems perspective.

In the logistic regression models, older siblings of children with ID were found to be less likely to be identified as hyperactive/inattentive compared to those siblings whose brother or sister did not have ID. In previous MCS research, data show that the children with ID in the sample were also more likely to be hyperactive (Totsika et al., 2011), and in this sample where the cohort children have an older sibling, 27.5% of children with ID are identified as having elevated hyperactive behaviours compared to 5.4% of those cohort children without ID. This contrast may help to explain the current findings. It is possible that primary caregivers may have completed the measure of the older siblings' hyperactivity in the context of the behaviour of their younger brother or sister with ID. Parents therefore may have been indicating that, relative to their brother or sister with ID, the older sibling was less hyperactive. To explore this further, and other potential explanations of this finding, in future research, it would be particularly useful to gather data from multiple informants (e.g., the older sibling themselves and possibly their class teacher). It is also possible that this finding may have been a Type I error due to multiple testing of the dataset, and this should be examined further in additional research.

How do findings from the present study help address the questions of whether, and how, siblings of children with ID might be at increased risk for behavioural and emotional problems? First, it is clear that these siblings are an at-risk group. The current population-based sample confirms this, and although the increased risk is 1.5—2 times that for other older siblings, only a minority of siblings of children with ID may experience (up to approximately 20% - see Table 2.3) elevated problems of behavioural or emotional adjustment. Second, the regression models suggest that a range of social and family factors are associated with sibling adjustment. Most of these putative risk factors (Table 2.1) also occur at higher levels in families of children with ID. However, given the cross-sectional design, we cannot distinguish how these factors may affect sibling adjustment. One hypothesis is that there is no effect of child ID on their siblings' behavioural and emotional adjustment but other social and family variables determine sibling adjustment. An alternative hypothesis is that growing up with a brother or sister with ID indirectly affects siblings' adjustment by directly increasing other risks (e.g., exposure to poverty, carers with psychological problems, and a brother or sister who also has behavioural and emotional problems). These alternatives should be explored

in future research, especially longitudinal research that can establish causal pathways for siblings' behavioural and emotional adjustment.

Existing literature has explored a number of other demographic variables (the siblings being of the same or different sex, sibling age, and the age difference between the older sibling and the child with ID) as correlates of siblings' behavioural and emotional adjustment (Cuskelly & Gunn, 2006; Ivey & Barnard-Brak, 2009; Martin & Horriat, 2012; Burke et al., 2012; Goudie et al., 2013; Walton, 2016). However, these factors did not emerge as significant predictors in the logistic regression models in the present study. This may have been because the data focused only on older siblings, or because these factors are more important within families of children with disabilities rather than for siblings generally. Existing population based studies exploring sibling differences included not just older siblings, but both older and younger siblings (Goudie et al., 2013; Emerson & Giallo, 2014; Neely-Barnes & Graff, 2011). Furthermore, the cohort children in this sample were all young. It is foreseeable that as children with ID age their behaviour may be perceived as more challenging and this may have a greater effect on family members, including siblings.

Although a strength of the current study is the population-based nature of the sample, a key limitation of the present study is that it only includes older siblings. There are some data to suggest that birth order may have an impact on various elements of sibling experience. Saxena (2015) highlighted that in adulthood, older siblings have been found to be more involved in care and suggest this may be in response to parental expectations of older siblings in childhood. There are also data to suggest that relationships between siblings and their brother or sister with developmental disabilities may be more 'positive' when they are the older sibling (Orsmond et al., 2009). Therefore, the current findings may not apply to the behavioural and emotional adjustment of younger siblings.

There are a number of limitations related to the nature of doing a secondary analysis, in that any analysis is limited by the variables made available. For example, it was not possible to identify if the older siblings themselves had ID or any other disability or genetic difference. Related to the population-based nature of the sample, the ID grouping variable is likely to include mostly children with mild to moderate

ID. Using a population based sample from a national birth cohort study meant that children with rare disorders or severe to profound ID would have been missed from sampling or assessment processes (because of very low population prevalence). Further research is needed to explore sibling behavioural and emotional adjustment when they have a brother or sister with severe ID and/or identified genetic syndromes. In addition, only one measure (the SDQ) was used to explore sibling outcomes. The SDQ is a screening measure for mental health rather than a more complete measure of sibling psychological adjustment. Future research should include a broader range of outcome measures for siblings. A final limitation is that the data in this analysis were based on primary carer reports of sibling adjustment and also of all other study variables. Thus, there is a problem of source variance. In future research, multi-informant methods, including sibling self-reports, are also needed (Kovshoff et al., 2017).

In terms of practical implications, policy makers and practitioners may want to concentrate support on siblings of children with ID who are considered at greater risk of other adversities – such as socio-economic hardships, high levels of primary carer psychological distress, and where their brother or sister with ID has significant behavioural and emotional problems. There is more work to be done on a structural level to address socio-economic inequalities or through specifically targeted family interventions to support these more-at-risk siblings and their families.

Tables

Table 2.1. Group differences for demographic and family factors for older siblings of children with or without ID (results weighted)

Demographic and family factors	Siblings of children without ID [95% CI]	Siblings of children with ID [95% CI]	χ^2	<i>p</i>
Older sibling male	51.2% [49.6, 52.7]	51.9% [45.3, 58.4]	0.05	.826
Household experiencing socio-economic deprivation	44.0% [41.8, 46.3]	77.6% [70.8, 83.3]	102.08	<.001
Siblings are same sex	50.0% [48.4, 51.6]	46.0% [36.5, 55.8]	0.78	.426
Single parent household	15.9% [14.8, 17.1]	28.9% [23.2, 35.4]	27.80	<.001
Main carer experiencing psychological distress	3.3% [2.9, 3.9]	6.7% [4.0, 10.9]	6.27	.013
Cohort child with or without ID having an SDQ total difficulties score in the 'abnormal' range	4.3% [3.6, 5.0]	27.8% [19.5, 38.0]	127.57	<.001
Primary respondent was natural parent of cohort member	99.6% [99.3, 99.8]	100.0% [100.0, 100.0]	.49	.953
Primary respondent was female	97.9% [97.4, 98.4]	97.8% [93.5, 99.3]	0.01	.928
	Siblings of children without ID mean (<i>SD</i>)	Siblings of children with ID mean (<i>SD</i>)		<i>p</i>
Number of children in the household	2.83 (1.48)	3.31 (1.26)		<.001
Age of older siblings	9.41 (3.47)	10.02 (3.29)		.004
Age of cohort member children	4.80 (0.58)	4.82(0.46)		.770
Age difference between older sibling and cohort member	3.68 (2.74)	3.85 (2.89)		.515
Age of primary respondents	35.21 (9.57)	33.39 (6.19)		.002
<i>Note.</i> df for each test = 1, 389 with the exception of: Age of cohort member children; Age difference between older sibling and cohort member; and Age of primary respondents df = 1, 387; Single parent household df = 2, 773; and Primary respondent was natural parent of cohort member df = 3, 1221.				

Table 2.2. SDQ mean scores for siblings of children with ID and without ID (results weighted)

SDQ score	Non ID mean (SE)	ID mean (SE)	ID mean difference [95% CI]	<i>t</i>	<i>p</i>	Cohen's <i>d</i>
Total behaviour problems	7.48 (.10)	8.98 (.49)	-1.50 [-2.50, -.51]	-2.97	.003	.19
Peer problems	1.42 (.03)	2.01 (.15)	-0.59 [-0.89, -0.29]	-3.85	<.001	.26
Conduct problems	1.54 (.03)	2.03 (.14)	-0.49 [-0.77, -0.21]	-3.46	.001	.22
Emotional symptoms	1.79 (.03)	1.93 (.19)	-0.14 [-0.53, 0.25]	-0.68	.495	.05
Hyperactivity/inattention	2.77 (.04)	3.05 (.17)	-0.29 [-0.63, 0.06]	-1.65	.101	.09
Prosocial	8.49 (.02)	8.31 (.12)	+0.18 [-0.06, 0.43]	1.48	.139	.09

Note. df for each test = 1, 389

Table 2.3. Group differences for elevated SDQ scores (results weighted)

	Siblings of children without ID 'abnormal' score [95% CI]	Siblings of children with ID 'abnormal' score [95% CI]	OR	OR 95% CI
SDQ total behaviour problems	8.9% [8.1, 9.7]	13.9% [9.4, 20.1]	1.65	1.04, 2.62
Peer problems	11.6% [10.8, 12.6]	20.9% [15.3, 27.8]	2.01	1.37, 2.95
Conduct problems	12.0% [11.0, 13.1]	19.2% [14.0, 25.8]	1.75	1.19, 2.57
Emotional problems	10.3% [9.6, 11.2]	12.2% [7.6, 19.2]	1.21	0.70, 2.10
Hyperactivity	7.1% [6.4, 7.8]	4.7% [2.4, 8.9]	0.65	0.33, 1.30
Prosocial	15.8% [14.9, 16.8]	18.8% [13.8, 25.2]	1.23	0.85, 1.79

Note. df for each test = 1, 389

Table 2.4. Logistic regression models showing correlates (odds ratios and 95% CIs) of elevated SDQ total behaviour problems score and sub-scores (results weighted)

	Elevated total problems score	Peer problems	Conduct problems	Prosocial behaviour	Hyperactivity/ inattention	Emotional symptoms
	OR [95% CI]	OR [95% CI]	OR [95% CI]	OR [95% CI]	OR [95% CI]	OR [95% CI]
Cohort child has intellectual disability	0.88 [0.41, 1.89]	0.94 [0.49, 1.77]	0.83 [0.42, 1.66]	0.88 [0.44, 1.76]	0.30 [0.10, 0.87]*	0.67 [0.34, 1.35]
Older sibling being male	2.03 [1.55, 2.65]***	1.74 [1.39, 2.17]***	1.59 [1.26, 2.00]***	2.23 [1.82, 2.73]***	2.93 [2.15, 4.00]***	0.80 [0.64, 1.00]*
Socio-economic deprivation	1.90 [1.44, 2.51]***	1.63 [1.27, 2.09]***	1.75 [1.39, 2.21]***	1.33 [1.11, 1.60]**	1.53 [1.13, 2.07]**	1.44 [1.12, 1.84]**
Cohort child - elevated SDQ total problems score	3.29 [2.17, 4.99]***	3.28 [2.30, 4.70]***	2.48 [1.65, 3.72]***	1.55 [1.00, 2.40]*	2.27 [1.35, 3.82]**	2.73 [1.86, 4.03]***
Single parent household	1.70 [1.23, 2.36]**	1.53 [1.14, 2.04]**	1.71 [1.31, 2.23]***	1.29 [0.99, 1.67]	1.76 [1.23, 2.51]**	1.38 [1.03, 1.87]*
Primary carer experiencing psychological distress	2.47 [1.46, 4.18]**	2.19 [1.27, 3.75]**	2.48 [1.56, 3.95]***	1.28 [0.80, 2.04]	1.64 [0.81, 3.28]	2.61 [1.61, 4.24]***
Older sibling age	0.69 [0.49, 0.98]*	0.89 [0.66, 1.20]	0.79 [0.60, 1.03]	0.95 [0.73, 1.24]	0.83 [0.58, 1.18]	0.77 [0.56, 1.06]
Number of siblings and cohort children in household	0.99 [0.82, 1.20]	0.98 [0.83, 1.16]	0.89 [0.76, 1.05]	0.84 [0.73, 0.97]*	1.10 [0.91, 1.34]	1.11 [0.90, 1.35]
Older sibling and cohort child being the same sex	0.85 [0.65, 1.11]	1.08 [0.85, 1.36]	0.97 [0.78, 1.21]	1.04 [0.84, 1.29]	0.92 [0.69, 1.21]	0.97 [0.78, 1.20]
Age difference between cohort child and older sibling	1.40 [0.99, 1.96]	1.07 [0.80, 1.45]	1.17 [0.90, 1.52]	0.93 [0.72, 1.21]	1.18 [0.84, 1.66]	1.35 [0.98, 1.86]

Note. * $p \leq 0.05$; ** $p < 0.01$; *** $p < 0.001$.

df for each predictor in the logistic regression models = 1, 386

Chapter 3: Study 2 - Supporting siblings of children with a special educational need or disability: an evaluation of Sibs Talk, a one-to-one intervention delivered by staff in mainstream schools²

² This study has been published in *Support for Learning* as follows: Hayden, N. K., McCaffrey, M., Fraser-Lim, C., & Hastings, R. P. (2019b) Supporting siblings of children with a special educational need or disability: an evaluation of Sibs Talk, a one-to-one intervention delivered by staff in mainstream schools. *Support for Learning*. 34, 4. 404-420. doi: 10.1111/1467-9604.12275. We would like to acknowledge and thank the school staff members and pupils for taking part in the evaluation of Sibs Talk. This study was conducted in collaboration with the UK charity Sibs, specifically Monica McCaffrey, a sibling of someone with IDD, and Clare Fraser-Lim. We would also like to acknowledge the administrative support provided by Louise Scott from Sibs, UK.

Chapter Three: Study Two

Chapter three (study two) sought to evaluate an intervention developed by the UK charity, Sibs. Our findings from the first empirical study (chapter 2) highlight that there are a group of siblings that may be in need of additional supports, and Sibs Talk is one way that this support may be provided. The intervention sought to support siblings of children with disabilities more broadly through a ten-session, manualized intervention. The intervention is unique, firstly, because it was a school-based intervention, and secondly, because it was a one-to-one intervention. We have identified no other studies evaluating interventions for siblings that were either one-to-one or school-based (Hartling et al., 2014; Tudor & Lerner, 2015; McKenzie Smith et al., 2018). The Embedded Systems Framework (Kovshoff et al., 2017) supports considering schools as part of the wider community that families and siblings interact with, this suggests that there may be value in considering the school-context as a source of support for siblings and families. This intervention and evaluation of it was embedded within a wider discussion about support for siblings and siblings' experiences at school. There has been relatively little research exploring siblings' school-based experiences, although population-based data from Goudie et al. (2013) indicated that siblings were more likely to have problems at school. A systematic review about school-based experiences of siblings of children with chronic illness described the psychological impact on schooling, decreases in school attendance, decreases in academic functioning, and issues with peer and teacher interactions (Gan et al., 2017). A recent small-scale survey found that siblings of autistic children had lower academic self-concept, sense of belonging at school, and worse behaviour problems than other adolescent siblings (Gregory et al., 2020). Cumulatively, these studies suggest that schools may be a possible context to provide support for siblings.

Abstract

A group often overlooked for specific supports in schools are siblings of children with a disability, special educational needs or a serious long-term condition (SEND). In this paper we review the current sibling research and we identify a lack of literature on interventions, particularly within a school context. We then present a description of Sibs Talk, an example of a new school-based intervention to support siblings. Sibs Talk is a ten-session, one-to-one intervention approach for schools to complete with Key Stage 2 children who have a brother or sister with SEND. Finally, we present an initial evaluation of the effectiveness of Sibs Talk using a pre and post evaluation format with a sample of 55 children from 11 schools. The data presented in this evaluation indicate that Sibs Talk may have contributed to positive outcomes for participating children.

Introduction

Schools have a responsibility to safeguard, ensure the well-being and enhance the education of the children in their care. A group often overlooked by UK schools are siblings of children with a disability, special educational needs or a serious long-term condition (hereinafter referred to as SEND). It has been estimated that approximately 7-17% of children are siblings of children with a chronic condition/disability (McKenzie Smith et al., 2018). Therefore, there are an estimated two to five siblings of children with SEND in the average UK classroom. The research evidence for sibling outcomes is rather mixed, with some studies indicating quite large negative differences in psychological well-being for siblings of children with SEND (Goudie et al., 2013), whilst other research indicates these well-being differences may be small and more likely due to indirect effects related to factors such as family socio-economic disadvantage (Emerson and Giallo, 2014; Neely-Barnes and Graff, 2011; Hayden et al., 2019a). Other research also indicates benefits and positive experiences of siblings of children with SEND (Mulroy et al., 2008).

More research needs to be done to understand sibling educational outcomes and experiences in school (Hastings, 2014; Kovshoff et al., 2017). There are some data to suggest siblings may be negatively affected educationally, with poorer functioning at school (Goudie et al., 2013). Whereas Chien et al. (2017) found that although siblings of children with autism had more behavioural problems and poorer attitude to school work, they had comparable academic achievements to children with a brother or sister without autism.

Generally, UK schools do little to support siblings of children with SEND as a specific target group. Some siblings will get support with well-being issues provided through general safeguarding and pupil support policies. However these are not targeted specifically for siblings of children with SEND. Furthermore this support may only materialise once concerns regarding siblings have already arisen – and there is a case for perhaps anticipating the needs of siblings – identifying and supporting this group before some siblings may encounter problems. A group that have recently gained more recognition from health and educational providers in the UK are young carers. The introduction of the Children and Families Act 2014 and the Care Act 2014 recognised and afforded new rights to young carers in England.

According to census data from 2001, there are at least 175,000 young carers under the age of 18 in the UK (Becker and Becker, 2008) with some reports indicating the figure could be nearer 700,000 (BBC, 2010). Although data suggest that most young carers are providing care for a parent, a large proportion are caring for their sibling – data varies, but would suggest between a quarter (Cheesbrough et al., 2017) and up to half of young carers are caring for a sibling (The Children’s Society, 2013). It remains unclear however, to what extent these young carers have a brother or sister with SEND, or whether they are helping with their younger ‘typically’ developing siblings. Therefore, schools should be considering sibling experiences when supporting young carers as well.

We have identified three systematic literature reviews that explored interventions for siblings of children with SEND (Hartling et al., 2014; Tudor and Lerner, 2015; McKenzie Smith et al., 2018). These systematic reviews highlight two factors making the present intervention unique. Firstly, the vast majority of interventions are delivered in hospital or other medical settings, community contexts, summer camps or in family contexts: it was not explicit that any interventions reviewed were based within the participating siblings’ school contexts. The second factor of note from these reviews is that, with very little exception, these interventions were peer group based rather than being multiple, one-to-one interventions between an individual sibling and an adult. Therefore, a one-to-one intervention delivered in siblings’ own schools may have unique potential benefits that are worthy of exploration.

Intervention Information and Basis

Sibs Talk is a one-to-one, manualised support intervention developed by the UK charity Sibs for pupils in Key Stage 2 who have a brother or sister with SEND. Sibs Talk consists of ten sessions spread over a school term aiming to improve siblings’ well-being and their engagement with learning (Sibs, 2018). The Sibs Talk intervention was informed by almost twenty years of the charity Sibs’ work. Knowledge developed through practice, working both directly and indirectly with siblings, indicated that many siblings were not identified in their school settings. Siblings’ needs can be overlooked leading to problems with well-being and progress at school. These children face various barriers to learning including disrupted sleep,

anxiety about their brother or sister's health, and less parental support with homework due to the demands of care. Sibs has worked with organisations across the UK to help them develop and run sibling support groups. These involve both recreational and discussion activities, providing siblings a space outside of the family home to meet other siblings, to share experiences and feelings as well as learn coping strategies. Over the past five years, fewer organisations have had the necessary funds or staffing levels to continue to run these groups in their local authorities. With this change in the funding climate, Sibs developed Sibs Talk, a one-to-one intervention for schools that would deliver some of the outcomes that were achieved through the support groups whilst responding to requests from school staff for a tool to support siblings. As most of these requests had been provided by primary schools, the Sibs Talk pilot was designed for Key Stage 2 pupils.

The content and format for Sibs Talk was based on the approach that Sibs teach professionals to use in sibling groups, with a key focus on acknowledging siblings' feelings and experiences, and facilitating discussion around coping strategies. A ready-to-use manualised tool was developed to minimise the time it would take for schools to run a one-to-one intervention with the hope that this would increase Sibs Talk participation. Design decisions were made through discussions with siblings, parents and school staff. These discussions led to decisions on the main topics covered in Sibs Talk, the number and length of the intervention sessions, which staff members would deliver the intervention, as well as the training needs of these staff. Once the Sibs Talk pupil activity booklet was written it was sent to a reader panel for feedback before the final version was produced. The reader panel consisted of a primary school head teacher, a SENCO, a school counsellor, a sibling support worker and an adult sibling. Each Sibs Talk pack consisted of a pupil activity booklet, a leaflet for parents about Sibs Talk, stickers, a certificate upon completion of the intervention, ethical documentation and the evaluation forms for staff and pupils to fill in (see Appendix Two, 2.1-2.12 for study documentation and ethics).

There are ten sessions in the intervention to be carried out during one school term. Each session lasts about 25 to 35 minutes. Most schools decided to complete sessions with siblings during scheduled lessons rather than at lunch time or after school. The staff member guides the sibling through an activity page in the booklet,

using the instructions in the Sibs Talk staff instruction manual. The first session starts with sharing basic information about each sibling's family and circumstances to help the sibling develop trust and rapport with the staff member leading the intervention. As the booklet progresses the sessions focus in more depth on their brother or sister's disability or condition, the sibling's feelings and experiences, and the issues that are challenging for them at home and school (see Figures 3.1 and 3.2). The sessions also look at the skills, knowledge and attributes that siblings have acquired and how their school can support them.

Prior to leading the intervention, staff attended a two-hour training session at their own school or at a host school nearby. The training focused on the micro skills required for listening to and acknowledging siblings' feelings. This involved a role play to practice how to respond to siblings' emotive questions and statements such as 'Will my brother die?' or 'I hate my sister'. Many school staff told us that their instinctive responses would be to explain things to the pupil or to help find a solution to the problem, so the training enabled staff to acknowledge siblings' feelings before taking any other action. Once the intervention was underway staff could contact Sibs staff for further advice if required. There were very few of these queries and most were to discuss support for specific siblings rather than for clarifications on delivering the Sibs Talk intervention.

Schools were recruited by approaching academy chains, individual schools and school staff in local authorities who had already shown an interest in sibling support or had a previous link with the charity. The Sibs Talk pilot information was also sent out through local Educational Psychology networks to recruit new schools. Sibs then identified and made connections with key people in local authorities who had access to local inter-school communication networks. Recruitment of schools was eased by linking Sibs Talk to the schools' existing policies and programmes on well-being and resilience, young carers, difference and diversity, anti-bullying, safeguarding and SEND support. Once schools agreed to take part in the pilot project and staff had participated in training, regular contact was maintained with the school for support, guidance and feedback.

Evaluation of Sibs Talk

The Participants and their Schools

The Sibs Talk Pilot was evaluated to help inform Sibs' future work with young siblings. In total, 55 evaluation questionnaires from 11 schools were returned and completed sufficiently to include in the evaluation. For eight of these schools, all children who completed the intervention also had the questionnaires completed and returned. The remaining three schools in the sample each returned four out of five of the possible completed questionnaires. Siblings were in Key Stage 2 and aged between seven and 11 years old (mean age = 9.18 years). There were slightly more female than male siblings in the sample (54.5% were female). In terms of ethnicity, 43.6% of the siblings were white-British and the three next largest ethnicity groups for siblings were Pakistani (23.6%), Black Caribbean (9.1%) and Black African (7.3%). Almost half of the siblings had a brother or sister with autism (49.1%), the rest of the siblings had a range of other disabilities including Down syndrome, hearing impairments or chronic medical conditions. In the sample, 36.4% of the children attracted Pupil Premium funding, and 36.4% of siblings spoke English as an additional language. Both of these factors occurred at a higher percentage than the UK school population average (GOV.UK, 2018). Although Sibs is a UK-wide charity and we have drawn on UK-wide data to understand siblings generally and understand the sample further, the sample of schools for this study were based in England only. There were 11 primary schools in total that returned evaluations for participating siblings. These schools included a faith school and an academy as well as schools from both rural and urban areas from across England, although with a particularly high uptake from schools in the South East of England.

Full institutional ethical approval (see Appendix Two, 2.1-2.12 for study documentation and ethics) was gained before 270 intervention packs were sent to 35 schools between March 2017 and February 2018. Schools voluntarily agreed to take part in Sibs Talk after receiving information from Sibs, UK. The Sibs Talk recruitment information was distributed by sharing information through local education networks, which vary greatly from place-to-place. Sibs expanded their existing school contacts to promote Sibs Talk by approaching Academy chains, individual schools, local Educational Psychology networks as well as school staff in Local Authorities that had shown an interest in siblings through previous contact with Sibs.

Schools were recruited and trained by Sibs staff before the intervention took place and the intervention was led by a school staff member, most commonly a learning mentor, teaching assistant, or SENCO. The intervention packs included the evaluation questionnaires as well as the relevant information sheets, consent forms and the ethical procedures form. The school provided parents and carers with information sheets about the study. Parents and carers were given the opportunity to withdraw their consent to their child taking part in the intervention and the evaluation. The participating children were given age-appropriate information on the study evaluation and provided assent to their answers being shared with Sibs and the researchers. Class teachers were asked to complete a measure on participating siblings. They were provided with a full information sheet and would only complete the measure if they consented to take part in the study. Sibs' main contact at the schools – a member of each school's senior leadership team – also signed a document confirming that their schools had followed the ethical procedures for the evaluation data collection. Full information and training was provided to relevant school staff members to assist them in following the ethical procedures. All pupils' names were removed from any evaluation documentation before being sent to Sibs. This ensured that the children's identities were kept anonymous from both Sibs and the researchers.

Measures and Methods

There were two questionnaires used which were both completed before the intervention and after the intervention. Firstly, the class teacher completed the Strengths and Difficulties Questionnaire with Impact Supplement (SDQ; Goodman, 1997; Goodman, 1999). This measures the behavioural and emotional well-being of the siblings. The items provide seven distinct scales: emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behaviour, total difficulties and an impact score.

Secondly, the participating children completed the 'How I Feel About My School' Questionnaire (HIFAMS; Ford, 2013; Allen et al., 2018). The HIFAMS was completed by the children during the first, and again, during the last intervention session. This questionnaire includes seven statements about school. For example, siblings are asked to respond to: 'When I am in the playground I feel...'. Children

then select an emoticon graphic to complete the statement to indicate feeling 'Happy', 'OK' or 'Sad'. These seven responses are then combined to create an overall score ranging from 0-14 with higher scores indicating the child is happier in school.

Basic socio-demographic information was provided by a member of the senior leadership team. Participating children also provided short, written comments to four prompts. As this activity was part of the child's personal work over ten activities, additional permission to share this information was sought from the sibling before they completed the activity – resulting in 31 siblings sharing these written responses for the evaluation.

Both the SDQ and HIFAMS questionnaires were analysed using paired-sample t-tests. This test provides the difference in the children's mean scores for the range of measures described above between two points: at the beginning of the Sibs Talk intervention and at the end of the Sibs Talk intervention. Content analysis was employed to examine the written responses from the siblings. Content analysis benefits from being a flexible, (Hsieh and Shannon, 2005) clear and systematic way of analysing written data (Seale and Tonkiss, 2012).

Findings from HIFAMS and SDQ questionnaires

The main results are presented in Figure 3.3. This shows the mean scores on each scale in the questionnaires, before and after Sibs Talk, for the whole sample of children involved in this evaluation. Across all measures, children had more positive scores at the end of the intervention period compared to the beginning. The mean scores pre and post intervention and also the effect sizes associated with the change in scores are summarised in Table 3.1. To test whether these changes were statistically meaningful, we compared the pre and post-intervention scores using paired samples t tests. The results of these tests revealed that the improvement in SDQ hyperactivity scores ($p < .001$) prosocial behaviours ($p = .002$); total SDQ difficulties ($p = .009$); and emotional problems ($p = .016$) were all statistically significant. The changes in the HIFAMS score and SDQ conduct problems, peer problems and overall SDQ Impact scores were not statistically significant.

Findings from the Siblings' Written Responses

Siblings also wrote short responses to four written prompts. One statement prompted ‘These people can help me with sibling stuff at school...’. Siblings then listed people they felt could help them. All 31 of the siblings referred to their teachers in this response.

There were a further three writing prompts which were analysed collectively, being coded and categorised. These prompts were: ‘I’ve learnt that...The activities have helped me because [and]...Something I want to tell people about being a sibling like me...’. There were 15 initial codes identified. These were then reduced, combined and clustered together to communicate four overarching categories: Learning and Understanding; Communication and Relationships; Coping Strategies; and Challenges and Responsibilities.

Learning and Understanding was identified as a category for 24 of the siblings. This included the siblings indicating that they had learnt new things about disability more generally. The siblings also highlighted learning about their brother or sister’s SEND specifically as well as learning ‘to be more open-minded’:

‘I know a lot about my sister. I learnt that people do listen to what I have to say. I learnt to be more open-minded’.

‘To learn about my feelings when I’m angry, sad, jealous, lonely, guilty, worried, embarrassed’.

‘I had learnt that other people had different conditions and how people can help you with it’.

Communication and Relationships were mentioned by 21 siblings. Furthermore, all 31 siblings could name individuals they could talk to about sibling issues in response to that specific prompt, with all siblings naming a teacher they could talk to about their sibling experiences. Some siblings wrote positive things about their brother or sister such as:

‘Me and my brother have a special bonding’.

‘I have a sister that’s autistic and that makes me proud’.

The sibling comments were generally written positively with regards to their sibling relationships, the selected quotes highlight the way in which these siblings

felt their relationships were unique and something to feel proud of. Some siblings wrote about their relationships with teachers, parents and organisations that could help. Siblings wrote about feeling more able to talk about their feelings and experiences, highlighting that they felt listened to:

'I don't have to keep things to myself. That it is not my brother's fault he is how he is. I have learnt that I can share things with you'.

'It's okay to talk about my brother. I know a lot more about my brother's disability. People want to listen to me and talk to me... It's helped me talk to my family and think about my family. It's been some special time for me... I need somebody to talk to. People need to explain things to me and talk to me. I need to think about my little brother'.

The sibling that wrote the above comment was able to articulate their needs and expectations of support from adults and themselves with regards to their sibling experiences. A combination of learning about SEND and feeling listened to may contribute to helping siblings feel less alone in their experiences, for example, one sibling wrote: 'There are millions of children who have disabled siblings like me'.

Coping Strategies were mentioned by 12 siblings in their written responses. This included learning how to manage, talk, or reflect on feelings and emotions as well as more practical tools when encountering challenging situations:

'I take a deep breath and count to ten and walk away'.

'I have a feelings box and when I don't want to say my feelings out loud I write in my feelings box'.

Challenges and Responsibilities were a category in 16 of the siblings' responses. This category involved a range of factors, such as physical and verbal aggression from their brother or sister, or difficulty communicating their feelings as siblings of a child with SEND.

'My brother's behaviour is very violent because he wants his own way all the time and gets really angry'.

[I have learnt] *'...how to deal with tough stuff such as keeping an eye on him all the time'.*

Those few siblings who wrote more negatively about their sibling relationships identified experiences of heightened physical aggression from their brother or sister with SEND. This category also encompasses some of the additional caring or supporting roles and responsibilities some siblings had at home. For their role in supporting their families, siblings seemed both positive and proud of these roles:

'I support him and help him learn'.

'There is not a medicine for autism. I'm proud of the things I do for my sister'.

These challenges or responsibilities were often phrased with a sense of acceptance of their brother or sister with SEND. For instance one sibling wrote: 'It's hard but I still love him'.

Conclusions

Although the evaluation results indicate some small, positive improvements for the participating siblings from the start of the intervention until the end of the intervention, a number of cautions when interpreting these data should be highlighted. Firstly, it must be remembered that this evaluation is not a randomised control trial, which has the benefit of reducing the effects of biases. There are numerous other factors which may have contributed to the resulting change in children's questionnaire scores from the first point the measures were administered before the intervention, and the second time point, at the end of the intervention. In addition, the teacher and child knew that they were going to or had been involved in Sibs Talk and so their responses may be biased.

In addition, the sample size is small especially given the larger number of intervention packs distributed. The schools that were able to allocate staff time to complete both the intervention and evaluation with pupils may not be representative of schools more generally. There is also likely a great deal of variation between the school staff members who led the intervention in terms of knowledge and experience of disability. The measures used in this study are also limited in scope. For example, the short written answers from the siblings in response to prompts indicate that some of the siblings had learned things about their brother or sister's SEND, special

educational needs or medical condition, and this development was not captured in the questionnaires. In addition, the second evaluation questionnaires were completed shortly after or during the final intervention. Therefore, whether improvements in scores would be maintained over time is indeterminable. Furthermore, the sibling comments were in response to very specific writing prompts, and were completed with the school staff member who led the intervention. This may have influenced the siblings' written responses.

Considerations for Further Study

Although these results indicate small improvements for the siblings taking part, a more robust evaluation would be necessary to decipher a more complete and confident picture of the impact and value of the Sibs Talk intervention. Ideally, this would take the form of a randomised control trial to reduce the risk of research bias in the evaluation. Any further evaluation would also benefit from a more representative sample, both in terms of socio-demographic factors, and also in terms of the range of conditions the siblings' brothers and sisters have. This would allow us to ask questions about how different sub-groups of siblings respond to the intervention. There may be some value in including other outcome measures, such as a parent report, whether siblings ask staff for help at school after completing the intervention, or a measure of the siblings' knowledge of disability given some of the more qualitative comments written by siblings. The HIFAMS measure had a lot of missing data due to children selecting multiple responses to indicate variability in their feelings. This may indicate that the guidance we provided staff members in assisting the siblings to complete the questionnaire needs to be improved, or perhaps indicates another measure easier for a non-researcher to administer would be more suitable.

Broader Considerations and Implications

School spending per pupil has recently fallen in real terms in England by about 8% (Institute for Fiscal Studies, Sibieta, 2018). Therefore the Sibs Talk pilot was developed and Sibs began recruiting schools to take part at a time when school funding was higher. Although Sibs noted that school ethos appeared to influence school participation in Sibs Talk, the main barrier for schools in deciding to take part or completing the intervention through to the end was staff time pressure due to

financial cuts. This made it difficult to allocate staff time to facilitate the Sibs Talk intervention.

The school recruitment and data collection process of this evaluation was time intensive, particularly for a small charity such as Sibs. Recruiting schools, training school staff, the administration processes and maintaining school involvement over many months involved significant staff time. Retention of schools proved challenging, as 270 intervention packs were sent to 35 schools, yet for this evaluation only 11 schools returned evaluations for 55 siblings in the timescale for these data to be analysed.

Sibs has explored the possibility of staff delivering Sibs Talk to pupils in small groups as a way of reducing the staff time involved, though this would significantly alter the intervention with the loss of the confidential one-to-one aspect of the work. It would also involve additional planning time to match pupils together so that they could progress through the intervention at the same pace. Other options for delivering Sibs Talk such as through staff in special schools or community organisations would remove the relationship between school staff and pupils being formed during the intervention which is one of the main elements of Sibs Talk.

Sibs originally hoped that following a successful pilot that Sibs Talk could be rolled out to many more primary schools across the UK, and then to secondary schools with a modified version of Sibs Talk for pupils in Key Stage 3. With the current funding situation for non-teaching staff in schools in the UK the charity acknowledges the significant challenges in getting more schools on board to deliver Sibs Talk.

In summary, the indicative evaluation of Sibs Talk suggests there may be small to moderate benefits for siblings who take part, and it is available as a tool for supporting siblings in schools that have the staff capacity to deliver it. A robust research evaluation needs to be conducted to understand the effects of Sibs Talk further. This would take the form of a randomized control trial, with a larger more representative sample of siblings, which would follow up with siblings both immediately after the intervention, and a year after the intervention concludes to test ongoing effects.

Tables and Figures

Table 3.1. Summary of Paired Sample T tests for HIFAMS and SDQ pre and post intervention measures.

	Pre Mean (<i>SD</i>)	Post Mean (<i>SD</i>)	Cohen's <i>d</i> (effect size)
HIFAMS	11.31 (1.98)	11.37 (1.98)	.03 (small)
SDQ emotional symptoms	2.69 (2.92)	2.00 (2.15)	.38 (small)
SDQ conduct problems	1.02 (1.70)	0.92 (1.68)	.07 (small)
SDQ hyperactivity	3.06 (2.79)	2.12 (2.49)	.55 (medium)
SDQ peer problems	1.62 (1.65)	1.34 (1.76)	.20 (small)
SDQ prosocial behaviours	7.63 (2.38)	8.39 (1.85)	.47 (medium)
SDQ total difficulties	8.44 (5.92)	6.40 (5.71)	.38 (small)
SDQ impact sub score	0.64 (.91)	0.53 (1.39)	.09 (small)

Note. For the HIFAMS and prosocial behaviours sub-score, higher scores indicate a more positive response for/about the sibling. For all other SDQ sub-scores and total difficulties score, lower scores indicate a more positive response.



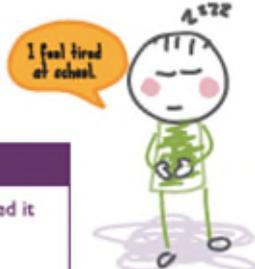
A booklet for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.

Figure 3.1. Front cover of the Sibbs Talk intervention manual

9. How school can help me

Siblings tell us that school can be difficult for lots of different reasons.

Colour in the circles if any of these things are difficult for you at school.

<input type="checkbox"/>	Struggling to get homework done	<input type="checkbox"/>	Feeling tired	<input type="checkbox"/>	Having to sort things for my brother or sister in school
<input type="checkbox"/>	Not being able to join in with things after school	<input type="checkbox"/>	Being bullied	<input type="checkbox"/>	Feeling worried or sad
<input type="checkbox"/>	Not having the right stuff with me for school	<input type="checkbox"/>	Being late or missing lessons		

I would like these things to happen

- Give me extra time to complete homework when I need it
- Help me to meet other siblings like me
- Let me check in at home when I am worried
- Keep an extra PE kit at school for me to use
- Don't tell me off if I am sometimes late

This week I can...

- Write down one thing which would really help me at school and give it to my teacher and my parent

tick

my idea...

School ideas

Figure 3.2. Session nine activity in the Sibs Talk intervention manual

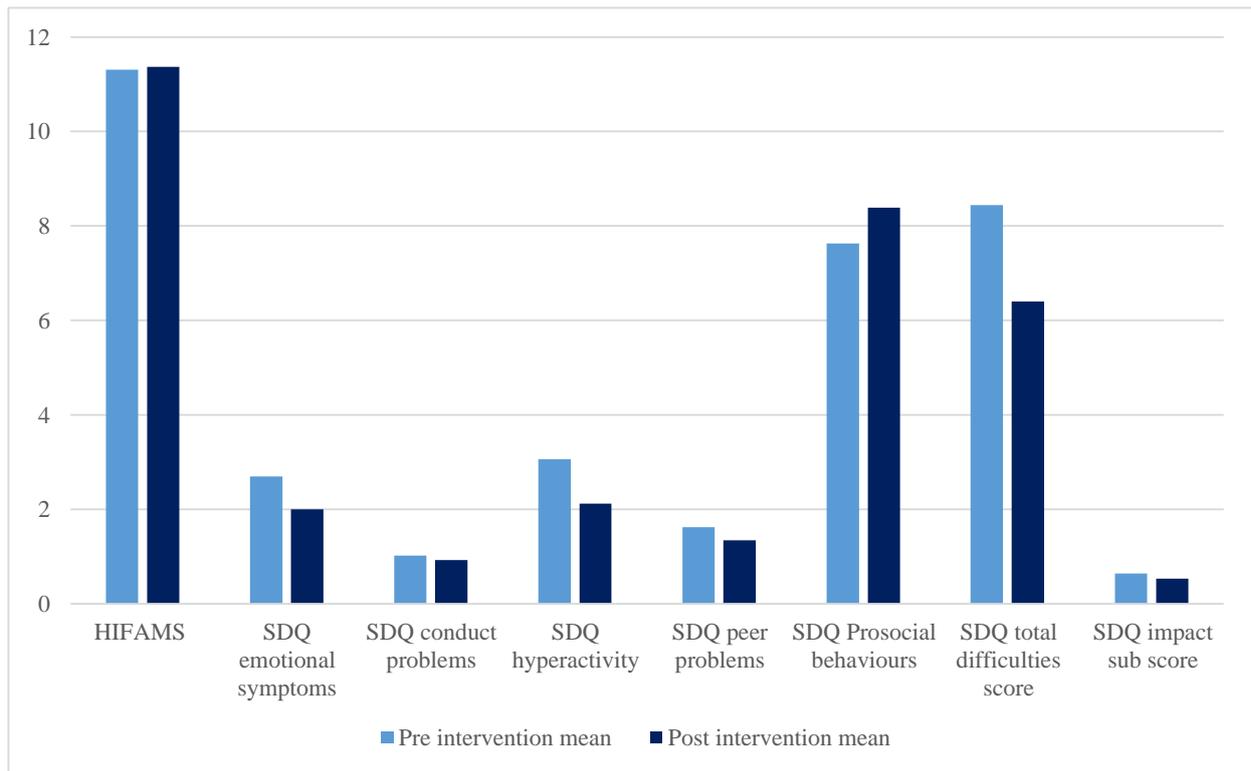


Figure 3.3. Pre and post intervention means

Chapter 4: Study 3 - Subjective poverty moderates the association between carer status and psychological outcomes of adult siblings of people with intellectual and developmental disabilities³

³ This study was designed in collaboration with the UK charity Sibs, specifically Clare Kassa and Frances Danylec, both of whom are also siblings of people with IDD. I acknowledge their support and expertise in designing and recruiting for this study. In addition, I am grateful to those adult siblings of people with IDD that provided feedback on an early draft of the survey used in this study. I am also grateful to Alison Baker from the Centre for Educational Development, Appraisal and Research at the University of Warwick, for her administrative support.

Chapter Four: Study Three

This is the only study out of the four empirical chapters that focused on adult siblings. As discussed in chapter one, empirical sibling studies are disproportionately about child siblings. Lifespan and developmental perspectives support the need to consider siblings' experiences throughout the lifespan. This study connects with all of the other empirical chapters by including measures about psychological outcomes (i.e. wellbeing and mental distress). It also includes a measure of physical health and quality of life, the latter of which is rarely considered in sibling research. The questions asked in this study go slightly further than those asked in the first empirical study (chapter two) as we include the moderating effects of SES and the level of independence of the person with IDD on the association between being a carer or not being a carer on sibling outcomes. This moves beyond simple main effects to consider interactions. Our findings from chapter two along with the empirical evidence about adult siblings presented in chapter one indicates that some siblings may be at risk of worse outcomes. This chapter sought to further understand, in adulthood, which specific groups of siblings may be at risk of worse outcomes. As in chapter two, we were able to include a range of covariates as well as various factors related to SES that were more developed than in much of the existing sibling research. As found in the child sibling study in chapter two, SES was an important covariate in understanding siblings' outcomes and so it was an important construct to include in multiple ways. This analysis approach of including a range of contextual factors as well as interaction terms allowed us to move beyond a negative narrative that assumes that siblings experiences worse outcomes simply because they have a brother or sister with IDD.

Abstract

Adult siblings are potentially important sources of care, support, advocacy, and friendship for their brothers and sisters with intellectual and developmental disabilities (IDD). Drawing on data about 911 adult siblings who completed an online national survey, we examined predictors and potential key moderators of siblings' mental distress, wellbeing, quality of life, and health outcomes. Compared to normative samples, adult siblings of people with IDD had worse mental distress (Cohen's $d = 1.20$), worse wellbeing ($d = .63$), worse quality of life ($d = .13$), and worse health ($d = .15$). Regression analyses indicated that carer status was not independently associated with sibling mental distress, wellbeing, and quality of life, although it was associated with poorer sibling health. Subjective poverty was associated with poorer sibling outcomes for all measures. Subjective poverty, but not the level of independence skills of the sibling with IDD, moderated the association between carer status and sibling mental distress, wellbeing, and quality of life. At low levels of subjective poverty, carer status was more strongly associated with sibling outcomes than at higher and average levels of subjective poverty. These findings indicate that some adult siblings may be at risk of worse mental distress, wellbeing, quality of life and health outcomes than other adults. Siblings experiencing higher levels of subjective poverty, with brothers and sisters with lower levels of independence, and siblings who are carers and also experiencing low levels of subjective poverty may be in need of specific supports.

Introduction

People with intellectual and developmental disabilities (IDD) are living longer lives than ever before (Coppus, 2013), with many people with IDD expected to outlive their parents' ability to provide care. Future planning for people with IDD often considers the potential of siblings to provide care and support for their brothers and sisters with IDD when parents are no longer able to (Lee & Burke, 2020). Understanding the psychological and health outcomes of adult siblings is important because siblings may provide care, support and friendship for their brothers and sisters with IDD (Lee & Burke, 2018), and will be more prepared for these roles if they are psychologically well and physically healthy. There is also a question about whether adult siblings may be at risk of worse outcomes themselves than other adults. This question would be consistent with family systems perspective, whereby family members are understood to influence and impact one another (White & Klein 2002; Smith-Acuña, 2011; Cox & Paley, 1997).

With the increased life expectancy amongst people with IDD, and the likelihood that siblings will take on caring later in life when their parents are no longer able to (Lee et al., 2019a), it is foreseeable that siblings may become more involved with the care of their brothers and sisters with IDD as they are facing more aged-related health problems such as physical health and mobility needs, behaviour changes, and dementia (Coyle et al., 2014). Sibling caregiving may also be unique, because siblings may anticipate and mentally prepare themselves for taking on a caregiving role from a young age. Furthermore, siblings may be more likely to be 'compound' carers (Lee et al., 2020), whereby they may care for their own children, their own parents, and their brothers and sisters with IDD, therefore managing multiple, distinct caregiving roles. Although positive aspects related to caregiving must be highlighted (Lee & Burke, 2018; Heller & Kramer, 2009), it is important to more fully understand the outcomes of sibling caregivers using validated measures to help us further understand whether sibling carers have support needs and what those might be.

Many existing studies about the outcomes of adult siblings have relied on small, convenience-based samples without a comparison group, making it difficult to fully contextualise findings. Some larger-scale studies have been published. For

example, Rai et al, (2018), drawing on a Swedish population-based cohort study of 223,842 participants, compared depression in young adults with and without autism. Part of their analyses drew on depression diagnoses in the cohort members' full and half siblings. They found that the full and half siblings of young adults with autism were at a slightly increased risk compared to the general population of having a depression diagnosis even when other socio-demographic factors (e.g. sex, age, family income) were considered (adjusted risk ratio for full-siblings = 1.37; adjusted risk ratio for half-siblings = 1.42). Taylor et al. (2008) drew on data from the Wisconsin Longitudinal Study to compare the outcomes of 268 adult siblings of people with mild ID, 83 adult siblings of people with mental illness, and 791 siblings of people without a disability. Taylor et al. found that although adult siblings of people with mental illness were more likely to have had a depressive episode in the past, the adult siblings of people with mild intellectual disability had similar outcomes to adult siblings of people without disabilities. These inconsistencies in findings relating to increased risk of negative outcomes for adult siblings may be associated with the geographical, generational, and disability-type differences in these two studies.

A literature review by Heller and Arnold (2010) further emphasised inconsistencies in research findings about adult siblings. In terms of the psychosocial outcomes of adult siblings, results were mixed, with eight studies highlighting positive psychosocial outcomes, and five studies showing more negative psychosocial outcomes for siblings. These differences in findings emphasise the need for analyses that consider questions about under what conditions some siblings experience worse or better outcomes. Such information might better inform appropriate support for adult siblings. This requires moving beyond analyses that explore direct effects of different risk factors to also consider moderated effects. We found two studies that used moderation analyses to explore the outcomes of siblings. Prino et al., (2019) found that gender moderated the association between sibling relationship quality and coping strategies. O'Neill and Murray (2016) found that none of their interaction terms moderated the association between being a sibling of someone with IDD and sibling depression or anxiety outcomes. Their moderation variables included: gender, socio-economic status (SES), age, and education levels.

Adult siblings may have differing outcomes based on whether they are a carer for their brother or sister with IDD. Much research about adult siblings as carers has concentrated on predicting future caregiving (Burke et al., 2012), future planning (Lee & Burke, 2020), factors associated with siblings caregiving (Lee et al., 2019b), and identifying support needs (Arnold et al., 2012). Less is known empirically about the outcomes of adult sibling carers as a specific sub-group of adult siblings, although Arnold et al., (2012) have speculated that the more involved in care siblings are, the more support needs siblings may have themselves. A systematic review by Lee and Burke (2018) identified sibling caregiving challenges, such as those related to emotional, economic, and physical needs; family conflict; and worries about challenges arising from ageing, and navigating services. One of the studies in the systematic review (Taggart et al., 2012), identified that over half of their sample ($N = 112$) of parent and sibling carers of people with IDD reported that caring was physically as well as emotionally demanding, with 40% self-reporting anxiety, and 31% self-reporting depression. A population-representative study about informal carers of people with intellectual disabilities (ID) explored the health, quality of life and impact of caring on carers of people with ID in comparison to other carers (Totsika et al., 2016). This sample is likely to include parental carers, but may also include adult sibling carers. Totsika et al. (2016) found that carers of people with ID had a similar quality of life to other carers, although they were at an increased risk of poorer health and were more likely to be struggling financially, which was related to poorer quality of life, health status, impact on health, and impact on personal life.

It is unclear in the sibling research literature what the psychological and health outcomes are for sibling carers specifically, and what is associated with the outcomes of sibling carers. Therefore, further research is needed to understand not just adult siblings' psychological and health outcomes, but also the outcomes of sibling carers. In addition it is important to consider what factors are associated with carer and non-carer adult siblings' outcomes, and how these factors interact.

Few studies identified in Lee and Burke's (2018) systematic review about caregiving roles of adult siblings included measures of socio-economic status (SES) and financial hardship (three of 29 studies included income). Of the three, Sonik et al. (2016) included and went beyond income to explore various aspects of socio-

economic status, including poverty, employment status, food security, receipt of benefits, and material hardship. Sonik et al. (2016) provided a descriptive comparison about adult sibling carers that lived with their brother or sister with IDD as the head of the household or partner of the head of the household ($n = 78$) and compared them to working-aged adults ($n = 64,555$) using USA, population-level, multi-wave data. They found that adult sibling carers were more likely to experience poverty and socio-economic hardships. Statistically significant comparisons found that sibling carers were more likely to be living 300% below the USA Federal poverty level, to not be employed in the past five weeks, to experience low or very low food security, and to receive public benefits compared to other adults of working age. Sonik et al. (2016) also explored education level and found that sibling carers were more likely to be less educated. Little work has been done in the caregiving or adult sibling literature about the association between SES and mental wellbeing and physical health outcomes. Measuring SES presents a methodological challenge, and moving beyond income and using cumulative measures of SES is an important way of capturing variations and nuances in SES amongst populations (Galobardes et al., 2007) and few sibling studies have explored this concept extensively. Further work is needed to not only incorporate a range of SES measures in sibling studies, but also to understand what role SES plays in the association between caregiving and sibling outcomes beyond exploring simple direct effects.

Another important factor that needs to be understood in relation to adult sibling outcomes and caregiving is the level of independence skills that the brother or sister with IDD has. Studies have explored whether the level of independence is associated with whether the sibling is already, or anticipates becoming, a caregiver for their brother or sister with IDD. These findings were mixed according to Lee and Burke's (2018) systematic review: they identified two studies finding that siblings were more likely to anticipate caring for their brothers and sisters with IDD when they had higher levels of independence (Krauss et al., 1996; Rimmerman & Raif, 2001), one study that found they were more likely to anticipate caring if their brothers and sisters with IDD had lower levels of independence (Seltzer et al., 1991), and one study found no statistically significant difference based on this factor (Heller & Kramer, 2009). Less considered in the existing sibling literature, is the level of independence of the brother or sister with IDD and what role sibling independence

skills play in the relationships between caregiving and the mental wellbeing and physical health outcomes of adult siblings. The non-sibling caregiving literature indicates that the more severe the disability of the person they are caring for, the worse outcomes caregivers will experience (e.g. parental caregivers of people with IDD, Seltzer et al., 2010). Those with lower levels of independence are likely to need more support and care.

Overall, there is a lack of existing evidence about the outcomes of siblings that are carers and non-carers, and that provide meaningful comparisons between adult sibling carers, adult sibling non-carers, and other adults. What other factors may be associated with adult sibling carer and adult sibling non-carer outcomes have also rarely been studied. The existing literature provides some support for considering how factors such as SES and the level of independence of the brother or sister with IDD may moderate the association between whether the sibling is a carer, and their psychological and health outcomes.

Therefore, we sought to explore the following research questions: (1) Does SES and the level of independence of the brother or sister with IDD moderate the association between sibling carer status and siblings' mental distress, wellbeing, quality of life, and health outcomes? (2) What are the mental distress, wellbeing, quality of life, and health outcomes of adult siblings, and how do these compare to other adults? For the first research question, drawing on the Double ABCX model (Hill, 1949; Hill, 1958; McCubbin & Patterson, 1983), we expected to find that both SES and the level of independence would moderate the association between siblings being a carer and their mental distress, wellbeing, quality of life, and health outcomes. Regarding the second research question, although findings in existing research vary, we expected to find that siblings may have slightly worse psychological scores (i.e. mental distress, wellbeing) compared to other adults (Rai et al., 2018). For quality of life, we were only able to compare our sibling sample to other informal carers, so we expected to find similar levels of quality of life to other carers (Totsika et al., 2016). For health, Hodapp et al., (2010) found that most adult siblings reported good health but we do not yet know how adult siblings' health compares to other adults.

Method

Participants

The sample consisted of 911 adult siblings of people with IDD. The mean age of adult siblings was 34.05 years ($SD = 12.78$, Range = 18 to 76 years). Adult siblings were predominately female (84.4% female; 14.9% male; 0.5% other; 0.1% missing). Data from the UK Indices of Multiple Deprivation (IMD), which measures relative deprivation of small geographic areas by utilizing national census data (Gill, 2015), indicated that 11% of the sample were living in the 20% most deprived neighbourhoods in the UK (13.4% missing). In terms of health, 30.5% of adult siblings indicated that they had a longstanding illness, disability, or infirmity. The largest ethnic group in the sample was White British ($n = 827$, 90.8%) followed by White Irish ($n = 17$, 1.9%), White Other ($n = 17$, 1.95%) and Asian Pakistani ($n = 7$, 0.7%). The largest religion group were 'No religion' (56.1%), followed by Christian (all denominations; 37.4%). The majority of the sample lived in England (87.8%), with 5.6% living in Scotland, 4.3% living in Wales, and 1.5% living in Northern Ireland (0.8% missing).

Adult siblings also provided background information about their brother or sister with IDD. The mean age of their brothers/sisters with IDD was 32.17 years ($SD = 13.88$, Range = 1 to 83 years), 49.0% were male, 32.9% were female; 1.0% were identified as 'other' and data were missing for 17.1%. Brothers/sisters with IDD were most commonly identified (non-exclusive categories) as having intellectual disability (47.9%), Autism (39.3%), Down syndrome (30.4%), or Cerebral Palsy (7.5%). Adult siblings were also asked to identify if their brothers/sisters with IDD had problems with various areas of physical health: 26.1% had visual impairments, 17.3% had hearing impairments, 34.9% had mobility problems, 40.5% had physical health problems, and 20.6% also had 'epilepsy/seizures'.

Measures

Adult Sibling Outcomes

Adult sibling mental distress was measured using the Kessler 6 (K6; Kessler et al., 2003). Participants were asked how often they have felt in the last 30 days: 'nervous', 'hopeless', 'restless', 'fidgety', 'so depressed that nothing could cheer you up' and whether everything was an 'effort and worthless'. Participants respond on a

five-point scale ranging from ‘all of the time’ to ‘none of the time’. These items are summed with scores ranging from zero to 24 and higher scores indicated more mental distress. We allowed one missing item to be replaced with mean estimation. Internal consistency (Cronbach’s α) for the K6 was .88 in the current sample.

Adult sibling wellbeing was measured using the Short-form Warwick Edinburgh Well-Being Scale (SWEMWBS; Stewart-Brown et al., 2009). This consists of seven, positively phrased items such as ‘I’ve been feeling optimistic about the future’ and ‘I’ve been feeling close to other people’. Participants were asked to rate these seven items on the following five-point scale based on their experiences in the preceding two weeks: ‘None of the time’, ‘Rarely’, ‘Some of the time’, ‘Often’, or ‘All of the time’. If there were any missing data for a participant, a raw score was not produced (Taggart et al., 2015). Raw scores were then transformed (Stewart-Brown et al., 2009) to facilitate cross-study comparisons, and to benefit from superior scaling properties. Final scores for the SWEMWBS were on a scale of seven to 35, with higher scores indicating better adult sibling wellbeing. Internal consistency (Cronbach’s α) for the SWEMWBS was .85 for the current sample.

Adult siblings’ quality of life was measured using a five-point scale of ‘Very good’, ‘Fairly good’, ‘Neither good nor bad’, ‘Fairly bad’ or ‘Bad’ in response to the following question: ‘If we were to define ‘quality of life’ as how you feel overall about your life, including your standard of living, your surroundings, friendships and how you feel day-to-day, how would you rate your quality of life?’ This question has been used in major UK national surveys such as ‘*The Survey of Carers in Households, 2009-2010*’ (2011).

Adult sibling health was measured by asking siblings to self-assess their ‘health in general’ on a five-point scale: ‘Very good’, ‘Good’, ‘Fair’, ‘Bad’ or ‘Very bad’ with higher scores indicating worse perceived health. This question has also been used in major national UK surveys including ‘*The Survey of Carers in Households, 2009-2010*’ (2011) and ‘*The Health Survey for England, 2018*’ (2020). A single item of self-reported health has been found to be a valid measure of overall general health (Idler & Benyamini, 1997).

Main predictor variables

Adult sibling participants were asked to identify (Yes or No) whether they were a carer for their brother/sister with IDD based on the following definition used nationally in the UK: ‘A carer is anyone, including children and adults, who looks after a family member, partner or friend who needs help because of their illness, frailty, disability, a mental health problem or an addiction and cannot cope without their support. The care they give is unpaid’ (NHS England, 2020).

Subjective poverty was measured by incorporating responses from two questions. Participants were asked how easily they could raise £2000 in an emergency and responded on a four-point scale whether they could ‘easily’ raise the funds, could do so by making ‘some sacrifices’, could do so by doing ‘something drastic’ or that they could not raise the money. This measure has been used in large-scale national surveys (Australian Institute of Family Studies, 2011). Participants were also asked to respond on a five-point scale to how well they were ‘managing financially these days’: ‘Living comfortably’, ‘Doing alright’, ‘Just about getting by’, ‘Finding it quite difficult’ or ‘Finding it very difficult’. This question was drawn from the Millennium Cohort Study (MCS, 2017), a UK nationally representative longitudinal study. Responses to these two items were z-transformed, then summed and divided by two (i.e. the number of items) to produce a single index of subjective poverty.

The level of independence in daily living of the brothers/sisters with IDD was measured using the Waisman Activities of Daily Living Scale (W-ADL; Maenner et al., 2013). Adult sibling respondents were asked to rate the level of independence their brother or sister with IDD had across 17 items on a three-point scale, ‘Independent or does on own’, ‘Does with help’, or ‘Does not do at all’ on statements including: ‘Making their own bed’ or ‘Doing errands, including shopping in stores’. Items were coded zero to two and summed so that total W-ADL scores ranged from zero to 34, with higher scores indicating that their brother/sister with IDD had higher levels of independence in their daily living. We made a small adjustment to the measure so that participants could also select ‘I do not know’ (ultimately coded as ‘missing’) to acknowledge that some siblings may not know about all aspects of their brother/sisters with IDD’s level of independence, particularly if they were not a carer. The original measure asks participants to answer about their son/daughter and we changed this wording to brother/sister. We allowed

a total W-ADL score to be calculated in cases where up to three items were missing, using mean estimation. Internal consistency (Cronbach's α) for the W-ADL was .94 in the current sample.

Procedure

Data were drawn from an online survey using Qualtrics™. The survey was designed through collaboration with Sibs, a UK based non-profit organization for brothers and sisters of disabled children and adults. Although three of the team that contributed to designing and scoping the survey were adult siblings, we also sought feedback from additional adult siblings that had not been involved in the survey design. Full ethical approval was granted by the University of Warwick's Humanities & Social Sciences Research Ethics Committee (see Appendix Three, 3.1-3.8, for study documentation and ethics). Initially, there were 927 participants who responded to the survey, but 13 cases were removed as they had consented but then not responded to any of the survey questions. To take part, participants had to be aged 18 years and over, had to live in the UK, have a brother or sister with IDD, and had to consent to take part in the research. Therefore a further three participants were removed because they were residing outside of the UK and our ethical approval only allowed us to recruit UK participants, resulting in an overall sample of 911 siblings. Participants were recruited from October 2019 until February 2020. Sibs led recruitment, advertising through their mailing list and through their social media networks (i.e. Twitter and Facebook). Sibs also contacted relevant IDD and family support organizations to encourage dissemination amongst further mailing lists and social media networks beyond Sibs' network. These advertisements and social media postings contained a link to the study information sheet and consent form on Qualtrics™.

Participants were asked to read the information sheet and to agree to each consent statement to progress onto the survey. The majority of participants that began the survey completed it until the end, with 715 of 911 participants completing the final survey item. The items in the survey that appeared to be associated with survey attrition were regarding financial circumstances of the adult siblings, which followed on from the main demographic questions. We explored group differences between those participants who did complete the income question and those who did

not, based on the demographic information that we had, but there were no statistically significant differences. In January 2020, we added two sentences to the survey reminding participants that they could skip questions and that their responses were anonymous but this did not improve retention following the financial questions.

Analysis procedure

All analyses were conducted using IBM SPSS Statistics 26. First, we explored whether each of the covariates were associated with the four sibling outcomes. Any that were not significantly associated with at least one of the four adult sibling outcomes were removed from further analyses. We then examined the remaining covariates to establish if any further variables should be removed due to multicollinearity, leading us to remove the age of the brother or sister with IDD as it was highly correlated with the age of the sibling ($r = .88$, $N = 754$, $p < .001$ two tailed). The age of their brother/sister with IDD was removed because it was slightly less associated with the sibling outcomes and because, conceptually, this study focused on the outcomes of the adult sibling rather than their brother/sister with IDD.

Moderated multiple regression models using Hayes' (2018) PROCESS macro (version 3.3; Model 2) were then conducted. We used mean centred product terms for the interactions. We included a range of covariates in our analyses based on commonly explored correlates in the existing sibling and broader caregiving literature: Age of both siblings (Heller and Kramer, 2009; Lee & Burke, 2020); whether the adult sibling participant was older or younger than their brother/sister with IDD (Egan & Walsh, 2001; Cleveland and Miller, 1977); the gender of both siblings (Orsmond and Seltzer, 2000; Sonik et al., 2016; Kraus et al., 1996; Cuskelly, 2016; Greemberg et al., 1999; Seltzer et al., 2005); whether they were the same or a different gender to one another (Heller and Kramer, 2009); whether the brother/sister with IDD had Down syndrome, autism (Orsmond & Seltzer, 2007; Hodapp & Urbano, 2007), or Cerebral Palsy; whether the adult sibling participant lived in the parental home and whether they lived with their brother/sister with IDD (Sonik et al., 2016); whether or not they had at least one non-disabled sibling in addition to their sibling with IDD (Burke et al., 2016; Lee et al., 2019b; Burke et al., 2012); whether they had one or more disabled siblings in addition to having a brother/sister with IDD (a broader caregiving study found that people caring for 2 or more people were

at a higher risk of poorer quality of life and health; Totsika et al., 2016); and additional measures of socio-economic position including having a degree-level qualification and living in a deprived neighbourhood (Sonik et al., 2016).

Results

Descriptive and Bivariate Analyses

Table 4.1 provides descriptive statistics and compares each of the four sibling outcomes – mental distress, wellbeing, quality of life, and health – with normative samples. Mean comparisons indicate that adult siblings had poorer scores on all four outcomes. For mental distress, adult siblings had higher mental distress scores (mean = 8.32, $SD = 4.94$, $N = 786$) compared to other UK adults (mean = 2.90, $SD = 4.08$, $N = 24761$). The difference between the two groups had a large effect size (Cohen's $d = 1.20$). Adult siblings also had worse wellbeing (mean = 21.25, $SD = 3.53$, $N = 770$) compared to other English adults (mean = 23.61, $SD = 3.90$, $N = 7196$). Therefore the adult siblings of people with IDD had worse wellbeing than other adults. The effect size for this difference was of a moderate size ($d = .63$). Although for quality of life and health, siblings of people with IDD had statistically significantly worse outcomes compared to other adults, the effect sizes for these group differences were small. Table 4.1 also compares adult siblings who were carers for their brothers/sisters with IDD with adult siblings who were not carers across the four sibling outcomes: mental distress, wellbeing, quality of life, and health. Across all four outcomes, sibling carers had statistically significantly worse outcomes compared to siblings that were not carers. However, the effect sizes for all four comparisons were small.

Table 4.2 explores the associations between each predictor/covariate and the four sibling outcomes. The tests presented in this table were used to decide which variables we would include as covariates in the moderated multiple regression models, and to check whether our test variables (carer status, SES, and level of independence of the brother or sister with IDD) were associated with the outcome variables. Variables that were statistically significantly associated with at least one of the four sibling outcome variables were retained in the moderated multiple regression models for all outcomes.

Table 4.3 provides a correlation matrix of the four adult sibling outcome variables: mental distress, wellbeing, quality of life, and health as well as the three main predictor variables: carer status, subjective poverty, and level of independence of the brother or sister with IDD. Each of the test variables were statistically significantly correlated with the four sibling outcome variables providing further support to include these variables in the final analysis models.

Moderated Multiple Regression Analyses

Tables 4.4 and 4.5 present the findings from the moderated multiple regressions for each of the four sibling outcomes. All models overall accounted a significant proportion of variance in the relevant outcome, with R^2 in the range 20-27%. Being a carer independently predicted sibling health ($\beta = .19$, $t(592) = 2.71$, $p = .007$), but not mental distress, wellbeing, or quality of life. Subjective poverty independently predicted all four sibling outcomes: mental distress ($\beta = 1.09$, $t(599) = 7.63$, $p < .001$), wellbeing ($\beta = -.88$, $t(586) = -8.48$, $p < .001$), quality of life ($\beta = .26$, $t(592) = 11.36$, $p < .001$), and health ($\beta = .20$, $t(601) = 7.82$, $p < .001$). The level of independence in daily living (WAD-L) of the brother/sister with IDD independently predicted sibling mental distress ($\beta = -.07$, $t(599) = -3.03$, $p = .003$), wellbeing ($\beta = .05$, $t(586) = 2.75$, $p = .006$), and quality of life ($\beta = -.01$, $t(592) = -3.31$, $p = .001$), but not sibling health ($\beta = .00$, $t(601) = .24$, $p = .813$).

Subjective poverty was found to moderate the association between status as a carer and mental distress ($F(1, 599) = 5.00$, $p = .026$, change $R^2 = .01$), wellbeing ($F(1, 586) = 6.76$, $p = .010$, change $R^2 = .01$) and quality of life ($F(1, 592) = 5.63$, $p = .018$, change $R^2 = .01$), but not health ($F(1, 601) = 1.39$, $p = .239$, change $R^2 = .00$). These three models showed the same pattern, whereby when adult siblings reported *low* levels of subjective poverty, status as a carer was associated with higher mental distress ($t = 2.40$, $p = .017$), lower wellbeing ($t = -2.20$, $p = .029$), and worse quality of life ($t = 2.16$, $p = .031$). At average and high levels of subjective poverty, the association between carer status and outcomes was statistically non-significant. There was no evidence that the level of independence of the brother/sister with IDD moderated the association between status as a carer and sibling outcomes.

No covariate was a statistically significant predictor in all four models. Younger siblings reported higher mental distress ($\beta = -.06$, $t(599) = -3.44$, $p = .001$),

better quality of life ($\beta = .01$, $t(592) = 3.10$, $p = .002$) and better health ($\beta = .01$, $t(601) = 3.25$, $p = .001$). Siblings that had multiple disabled siblings reported worse quality of life ($\beta = .23$, $t(592) = 2.07$, $p = .039$) and worse health ($\beta = .25$, $t(601) = 2.08$, $p = .038$) than siblings that only had one disabled sibling (i.e. only their brother or sister with IDD). Siblings with degree-level qualifications reported higher wellbeing than siblings without degree-level qualifications ($\beta = .84$, $t(586) = 2.51$, $p = .012$) and better health ($\beta = -.16$, $t(601) = -1.97$, $p = .049$). Siblings who lived with their brother or sister with IDD reported worse quality of life than siblings that did not live with their brother or sister with IDD ($\beta = .25$, $t(592) = 2.47$, $p = .014$). Siblings who had a brother or sister with Down syndrome compared to other conditions reported lower mental distress ($\beta = -1.26$, $t(599) = -2.88$, $p = .004$), higher wellbeing ($\beta = 1.31$, $t(586) = 4.17$, $p < .001$), and better health ($\beta = -.20$, $t(601) = -2.58$, $p = .010$). Siblings who had a brother or sister with Cerebral palsy compared to other conditions reported worse health ($\beta = .37$, $t(601) = 2.92$, $p = .004$).

Discussion

Overall (Table 4.1), adult siblings had worse scores in comparison to normative samples across all four measures of mental distress, wellbeing, quality of life, and health, although the effect sizes for quality of life and health were small. Like Rai et al., (2018) adult siblings reported worse mental distress than other adults. For health, Hodapp et al., (2010) found that most adult siblings reported good health. We also found that most siblings reported good health, but it was slightly worse than other adults, although with a small effect size. We also explored the mental distress, wellbeing, quality of life, and health outcomes of adult siblings who were and were not carers (Table 4.2). Siblings with carer status had slightly worse outcomes than siblings without carer status, with small effect sizes across the sibling outcomes. However, once we controlled for other covariates and moderating variables using moderated multiple regression (Tables 4.4 and 4.5), carer status, independent of all other variables, was only a statistically significant predictor for one outcome; sibling health.

Turning to our main analyses, there was evidence of a moderating effect of subjective poverty between carer status and adult sibling outcomes (mental distress, wellbeing, and quality of life). Subjective poverty was a statistically significant

variable in each of the four moderated multiple regression models. In addition, the interaction term between subjective poverty and carer status was also statistically significant in three of the models: mental distress, wellbeing, and quality of life. In each model, we found that where adult siblings had low levels of subjective poverty, carer status was associated with siblings' mental distress, wellbeing, and quality of life. We expected to see that carer status would be associated with poorer sibling outcomes at *high* levels of subjective poverty because we hypothesised that there may be a 'pile up' (Double ABCX model) or cumulative stressors acting in concert. Sibling participants with high levels of subjective poverty were at a heightened risk of worse outcomes across all four models. This is perhaps indicative of the ways in which deprivation and poverty are risk factors for worse outcomes more generally (Skapinakis et al., 2006; Connolly et al., 2000; Lakshman et al., 2011). Therefore, it may be that the stresses related to poverty were so impactful in the lives of the sibling participants experiencing higher levels of poverty that the stresses of being a carer had less additional impact.

The independent associations of the level of independence of the brother or sister with IDD were statistically significant in the mental distress, wellbeing, and quality of life models, but not in the health model. Siblings that had brothers or sisters with IDD with higher levels of independence had slightly less mental distress, better wellbeing, and better quality of life. We expected that siblings having a brother or sister with IDD and higher levels of independence would have better outcomes, because the types of care and support that siblings may provide for their brothers and sisters with IDD may not be as extensive, and therefore may have less of an impact on the siblings. No evidence was found that the level of independence of the brother/sister with IDD moderated the association between carer status and the sibling outcomes. This was contrary to our initial hypothesis, as we expected that siblings that were carers of a brother or sister with IDD who had lower levels of independence may be required to undertake more intensive levels of care, and that this may have more of an impact on their mental and physical health. Perhaps this is related to social welfare available in the UK for disabled people with lower levels of independence. It could be that siblings with brothers and sisters with IDD and lower levels of independence are receiving additional state-funded care and support that means the amount or the type of care that these siblings provide is of a different

nature, or less intense, than we might have assumed. Unfortunately, we did not include questions about the supports and services the individuals with IDD were receiving and so cannot examine this hypothesis.

The following covariates were statistically significant in at least one model: age of sibling, multiple disabled siblings, sibling educational qualification level, sibling living with their brother or sister with IDD, the brother or sister with IDD having Down syndrome, or having Cerebral palsy. Interestingly, autism was not statistically significant in any of the four outcome models. Existing studies have explored the outcomes of siblings of people with Down syndrome in comparison to the siblings of people with autism (Orsmond & Seltzer, 2007; Hodapp & Urbano, 2007) and have found evidence of the Down syndrome advantage, whereby family members of people with Down syndrome fair better than family members of people with other conditions. Population-based data about child siblings has also found evidence of the Down syndrome advantage (Marquis et al., 2020). We have also found evidence of the Down syndrome ‘advantage’ in the models measuring siblings’ mental distress, wellbeing, and health, but not quality of life. This suggests that although we can conceptualise a Down syndrome ‘advantage’ for adult siblings, it would not be appropriate to conceptualise autism as a risk factor for worse sibling outcomes, supporting a need to resist a ‘negative narrative’ (Hastings, 2016) of the effect having a family member with IDD may have on an individual.

Limitations

Most of the limitations for this study stem from the use of a survey study design and convenience sampling. Data for this study were from cross-sectional data only. Longitudinal data would be required to understand these associations further. All data were from sibling self-report data. We have no diagnostic information from relevant practitioners about the sibling (e.g. mental health diagnoses) or their brother or sister with IDD (e.g. to confirm they have an autism diagnosis). Our sample is also not likely to be representative of the UK population. We disproportionately recruited white, female, and well-educated participants and therefore we are unlikely to be able to generalise these findings to all UK siblings. We recruited through the UK charity Sibs, and we asked other disability support organisations to share the survey advertisement as well. Therefore, our participants were likely accessing or

receiving communication from support organisations. People that take part in IDD sibling research are also more likely to self-identify as ‘siblings’, and perhaps feel more affected by their experiences as siblings than other siblings.

A problem with caregiving research, and sibling caregiving research specifically, is that caregiving is not always defined for participants (Lee et al., 2019a; Lee & Burke, 2018) and where there is a definition of caregiving provided, it varies across studies (Lee & Burke, 2018). Although our study did provide a UK national definition of informal caregiving to guide participants, respondents were only able to select ‘Yes’ or ‘No’ in response to whether they felt the definition applied to them. Our binary definition is arguably ‘overly simplistic’ (Lee et al., 2018: 126). Other studies have probed the concept of caregiving further, exploring the concept as more of a continuum rather than as a binary construct (Lee et al., 2019b; Lee et al., 2018).

Only about 20-27% of the variance was explained by the moderated multiple regression analyses and our effect sizes were small. This indicates that there are other factors explaining siblings’ outcomes, and these may not be related to sibling status or carer status.

Future research directions

Longitudinal data are required to understand sibling outcomes further. Longitudinal data about adult siblings would help us understand how their experiences change over time. For example, data could explore how siblings’ outcomes fluctuate based on how their carer role, measured on a continuum, fluctuates. Longitudinal data would also allow for wider analysis potential, such as modelling reciprocal effects that explore how the outcomes of siblings are related to the outcomes of their brothers and sisters with IDD. Other longitudinal analyses could explore indirect effects using mediation analyses, to consider, for example, whether the association between carer status and siblings’ outcomes is mediated by the quality of the sibling relationship. Our study does emphasise the importance of looking beyond main effect predictors to moderated effects. Had we not tested for moderated effects, we would have incorrectly concluded that carer status was not important in relation to adult sibling outcomes once other factors have been

controlled. However, carer status was important but under certain conditions of subjective poverty.

Research generally, including family and sibling research such as the present study, consistently fails to recruit participants from marginalised communities. It is imperative that we develop effective ways of meaningfully involving people from marginalised communities in our work. Research studies by Sonik et al. (2016) and Richardson & Stoneman (2019) have been important in helping us understand race and ethnicity further in sibling research. Further research is needed to understand how ethnicity may affect siblings' experiences. We also under-recruited brothers and siblings with lower SES in this study, and this is common in sibling research. Further work should be done to also explore the experiences of brothers of people with IDD as a specific group of siblings as well as siblings with low SES. The definition and conceptualisation of carer status in the adult sibling research literature needs further development (Lee et al., 2019b; Lee et al., 2018; Lee & Burke, 2018) to consider carer status as a fluctuating continuum rather than as a binary definition. To more fully understand the range of factors associated with adult sibling outcomes, future research should also explore additional predictors at the individual, family, social network, community, and societal levels (cf. Kovshoff et al., 2017).

Practical and clinical implications

A conceptual issue with this study is that by exploring sibling outcomes, and finding that siblings have worse outcomes than other adults, we are implicated in advancing a 'negative narrative' (Hastings, 2016) about the effect of having a brother or sister with IDD. However, understanding more clearly the factors associated with the outcomes of a group that may potentially need further support, has importance and value. This is particularly so for siblings with carer status, who are relied on to support and care for their brothers and sisters with IDD. The current data suggest that some siblings may be in need of supports and interventions, particularly related to contextual factors such as for those siblings experiencing socio-economic hardships. Siblings that have a brother or sister with IDD and *lower* levels of independence may also require more support. Our data also suggest that siblings that have low levels of subjective poverty but that are carers may benefit from specific supports as well. Further research is necessary to understand this

finding further and consider why caring was associated with poorer outcomes for siblings with low levels of subjective poverty. Future research may consider including in analyses factors related to how siblings perceive the way that caring impacts their lives (i.e. carer 'burden' measures). The mental distress and wellbeing outcomes had large and medium effect sizes when compared with other adults using national data, whereas the effect sizes in these comparisons remained small for quality of life and health. Therefore, any interventions and supports might focus particularly on psychological outcomes related to mental distress and mental wellbeing.

Tables

Table 4.1. Sibling outcomes compared with normative samples and group comparisons between being a caregiver or not with sibling outcomes.

	Current sample mean (<i>SD</i>)	<i>t</i>	Normative mean (<i>SD</i>)	Cohen's <i>d</i>	Carer mean (<i>SD</i>)	Not a carer mean (<i>SD</i>)	<i>t</i>	Cohen's <i>d</i>
Mental distress	8.32 (4.94)	30.79***	2.90 (4.08) ¹	1.20	8.84 (4.96)	7.58 (4.78)	-3.47**	.26
Wellbeing	21.25 (3.53)	-18.51***	23.61 (3.90) ²	.63	20.95 (3.53)	21.59 (3.58)	2.41*	.18
Quality of Life	2.06 (.85)	3.71***	1.94 (.91) ³	.13	2.16 (.85)	1.93 (.82)	-3.72***	.28
Health	2.09 (.89)	4.55***	1.95 (.95) ⁴	.15	2.20 (.92)	1.93 (.84)	-4.20***	.31

Note.

* $p < .05$, ** $p < .01$, *** $p < .001$;

Comparison from the following datasets: ¹*NCHS, 'National Health Interview Survey, 2018'* (2020); ²*Health Survey for England data, 2011* (Warwick Medical School, 2020); ³*The Survey of Carers in Households, 2009-2010'* (2011); ⁴*The Health Survey for England, 2018'* (2020).

Sibling sample for each outcome: Mental distress $N = 786$; Wellbeing $N = 770$; Quality of Life $N = 777$; Health $N = 791$.

Table 4.2. Associations between potential covariates and outcomes.

	Mental distress		Wellbeing		Quality of Life		Health	
	<i>t</i>	<i>df</i>	<i>t</i>	<i>df</i>	<i>t</i>	<i>df</i>	<i>t</i>	<i>df</i>
Multiple non-disabled sibs	.03	745	.38	731	-.09	736	-1.86	705
Multiple disabled sibs	-.57	747	.40	733	-1.93	738	-2.19*	750
Sib is older/younger	1.00	723	-.05	709	-.65	714	-.78	726
Sib gender	-1.28	165	.71	764	1.11	771	-2.67**	785
IDDsib gender	-.48	741	-1.34	727	-1.13	598	-1.26	744
Same/different gender	.00	750	1.43	736	1.90	679	.53*	753
IMD bottom quintile	-3.64***	110	2.07*	678	-2.22*	110	-2.86**	112
Sib degree education	.461***	782	-3.87***	766	2.29*	260	2.94**	285
Sib lives with parents	-4.37***	782	3.39**	766	-1.58	774	.30	787
Sib lives with IDDsib	-4.23***	781	.38**	765	-2.57*	772	-.66	786
IDDsib autism	-2.04*	784	2.16**	768	-1.73	775	-.60	789
IDDsib Down syndrome	4.03***	784	-5.47***	512	3.12**	775	4.31***	630
IDDsib Cerebral Palsy	-1.93	784	3.71***	81	-3.09**	775	-2.99**	76
	Pearson's	<i>N</i>	Pearson's	<i>N</i>	Spearman's	<i>N</i>	Spearman's	<i>N</i>
	r		r		r		r	
Sib age	-.18***	786	.07	770	.03	777	.08*	791
IDDsib age	-.15***	751	.06	737	-.01	742	.08*	754
Subjective poverty	.34***	764	.34***	749	.42***	756	.33***	769
Independence IDDsib	-.15***	749	.16***	735	-.16***	740	-.08*	752

Note.

* $p < .05$, ** $p < .01$, *** $p < .001$.

Table 4.3. Correlation matrix of dependent variables and main predictors

	1	2	3	4	5	6	7
1. Mental distress	-						
2. Wellbeing	-.72***	-					
3. Quality of Life	.59***	-.63***	-				
4. Health	.43***	-.44***	.51***	-			
5. Sibling is a carer	.13**	-.09*	.14***	.15***	-		
6. Subjective poverty	.34***	-.35***	.43***	.35***	.12**	-	
7. Independence of IDD sib	-.15***	.16***	-.16***	-.09*	-.18***	-.07	-

Note.
 * $p = <.05$, ** $p = <.01$, *** $p = <.001$.

Table 4.4. Moderated multiple regressions for Sibling Mental Distress and Wellbeing

	Mental Distress (K6) ^a						Wellbeing (SWEMWBS) ^b					
	β	SE	<i>t</i>	<i>p</i>	95% LLCI	95% ULCI	β	SE	<i>t</i>	<i>p</i>	95% LLCI	95% ULCI
Constant	9.80	1.53	6.40	<.001	6.79	12.81	21.80	1.10	19.87	<.001	19.64	23.95
Sibling is a carer	0.42	0.39	1.08	.282	-0.34	1.17	-0.13	0.28	-0.45	.652	-0.67	0.42
Subjective poverty	1.09	0.14	7.63	<.001	0.81	1.37	-0.88	0.10	-8.48	<.001	-1.08	-0.67
Carer x Poverty	-0.62	0.28	-2.24	.026	-1.17	-0.08	0.52	0.20	2.60	.010	0.13	0.92
Independence of IDD sib	-0.07	0.02	-3.03	.003	-0.12	-0.03	0.05	0.02	2.75	.006	0.01	0.08
Carer x Independence	0.02	0.05	0.44	.659	-0.07	0.11	-0.03	0.03	-1.05	.294	-0.10	0.03
Model covariates												
Sib age	-0.06	0.02	-3.44	.001	-0.09	-0.02	0.00	0.01	-0.21	.837	-0.03	0.02
Multiple disabled sibs	0.20	0.68	0.30	.768	-1.13	1.53	-0.23	0.49	-0.47	.641	-1.18	0.73
Sib gender	0.80	0.53	1.51	.132	-0.24	1.84	-0.34	0.38	-0.88	.377	-1.10	0.42
Same/different gender	-0.28	0.38	-0.74	.457	-1.03	0.46	-0.27	0.27	-1.00	.319	-0.81	0.26
Neighbourhood deprivation	0.78	0.57	1.37	.172	-0.34	1.90	0.09	0.40	0.21	.832	-0.71	0.88
Sib education	-0.82	0.47	-1.77	.078	-1.74	0.09	0.84	0.34	2.51	.012	0.18	1.50
Sib lives with parents	-0.26	0.61	-0.42	.673	-1.46	0.94	0.21	0.44	0.48	.630	-0.66	1.09
Sib lives with IDD sib	0.86	0.63	1.37	.171	-0.37	2.09	-0.77	0.45	-1.71	.088	-1.66	0.12
IDD sib autism	-0.36	0.42	-0.86	.388	-1.19	0.46	0.15	0.30	0.49	.622	-0.45	0.75
IDD sib Down syndrome	-1.26	0.44	-2.88	.004	-2.12	-0.40	1.31	0.32	4.17	<.001	0.70	1.93
IDD sib Cerebral Palsy	-0.29	0.71	-0.42	.678	-1.68	1.09	-0.35	0.51	-0.68	.495	-1.36	0.66

Notes.

Model summaries (all predictors to Y):

^a $F(16, 599) = 9.48, p < .001, R^2 = .20, N = 616;$

^b $F(16, 586) = 10.03, p < .001, R^2 = .22, N = 603.$

LLCI: Lower Level of Confidence Interval; UCLI: Upper Level of Confidence Interval.

Table 4.5. Moderated multiple regressions for Sibling Quality of Life and Health

	Adult Sibling Quality of Life ^a						Adult Sibling Health ^b					
	β	SE	<i>t</i>	<i>p</i>	95% LLCI	95% ULCI	β	SE	<i>t</i>	<i>p</i>	95% LLCI	95% ULCI
Constant	2.00	0.25	8.07	<.001	1.51	2.49	1.60	0.27	5.84	<.001	1.06	2.14
Sibling is a carer	0.04	0.06	0.61	.543	-0.09	0.16	0.19	0.07	2.71	.007	0.05	0.32
Subjective poverty	0.26	0.02	11.36	<.001	0.22	0.31	0.20	0.03	7.82	<.001	0.15	0.25
Carer x Poverty	-0.11	0.05	-2.37	.018	-0.20	-0.02	-0.06	0.05	-1.18	.239	-0.16	0.04
Independence of IDD sib	-0.01	0.00	-3.31	.001	-0.02	-0.01	0.00	0.00	0.24	.813	-0.01	0.01
Carer x Independence	0.01	0.01	1.47	.142	0.00	0.03	0.01	0.01	1.38	.167	0.00	0.03
Model covariates												
Sib age	0.01	0.00	3.10	.002	0.00	0.01	0.01	0.00	3.25	.001	0.00	0.02
Multiple disabled sibs	0.23	0.11	2.07	.039	0.01	0.44	0.25	0.12	2.08	.038	0.01	0.49
Sib gender	-0.10	0.09	-1.21	.225	-0.27	0.06	0.17	0.09	1.82	.069	-0.01	0.36
Same/different gender	-0.11	0.06	-1.83	.068	-0.23	0.01	-0.06	0.07	-0.86	.388	-0.19	0.07
Neighbourhood deprivation	-0.02	0.09	-0.18	.857	-0.20	0.16	0.14	0.10	1.41	.158	-0.06	0.34
Sib education	-0.12	0.08	-1.56	.120	-0.27	0.03	-0.16	0.08	-1.97	.049	-0.33	0.00
Sib lives with parents	-0.03	0.10	-0.35	.730	-0.23	0.16	-0.14	0.11	-1.27	.203	-0.36	0.08
Sib lives with IDD sib	0.25	0.10	2.47	.014	0.05	0.45	0.21	0.11	1.90	.057	-0.01	0.43
IDD sib autism	0.03	0.07	0.43	.671	-0.11	0.16	0.00	0.08	0.01	.988	-0.15	0.15
IDD sib Down syndrome	-0.11	0.07	-1.50	.135	-0.25	0.03	-0.20	0.08	-2.58	.010	-0.36	-0.05
IDD sib Cerebral Palsy	0.11	0.12	0.92	.360	-0.12	0.33	0.37	0.13	2.92	.004	0.12	0.62

Notes.

Model summaries (all predictors to Y):

^a $F(16, 592) = 13.390, p < .001, R^2 = .27, N = 609;$

^b $F(16, 601) = 9.43, p < .001, R^2 = .20, N = 618.$

LLCI: Lower Level of Confidence Interval; UCLI: Upper Level of Confidence Interval.

Chapter 5: Study 4 - Behavioural adjustment of children with intellectual disability and their siblings is associated with their sibling relationship quality⁴

⁴ I acknowledge the support of Tom Bailey, who provided guidance on my analyses for this chapter.

Chapter Five: Study Four

Data from this final thesis study were derived from the first wave of the 1,000 Families Study,⁵ the largest UK sample of families of children with ID (Hastings et al., 2020). We derived a sub-sample of 500 sibling dyads, one with ID, and one sibling without IDD. Having such a large sample allowed us to use more advanced data modelling techniques, in this case, structural equation modelling. This allowed us to ask more interesting questions of our data in this chapter, such as allowing us to consider reciprocal associations at the sibling sub-system level. The studies in this thesis have thus far treated psychological constructs as the outcome variables and have, for the most part, considered direct effects. The prior chapter, chapter four, went a step further than direct effects, and examined interaction terms. The present chapter, my final empirical study, examined reciprocal effects, testing some of the sibling-dyad level conceptualizations in family systems theory. We included the psychological constructs, in this case behavioural and emotional outcomes, as the dependent variables, with sibling relationships as the main outcome and focus of the analyses.

In chapter one, I highlighted the importance of sibling relationships and included a discussion about the negative narrative and assumption made about siblings, that they are somehow negatively affected by having a brother or sister with IDD. This conceptualization oversimplifies the reciprocal effects that siblings have on one another. The empirical studies I presented in chapters two, three, and four arguably fall into this trap, by focusing on the siblings' psychological outcomes. This chapter, my last empirical study, began with the assumption that sibling relationships where one has an ID are similar to sibling relationships where neither have a disability, whereby both siblings' 'positive' and 'negative' behaviours will be associated with their relationships in both 'positive' and 'negative' ways. The intention of hypothesizing and conceptualizing sibling experiences in this way was

⁵ A description of the participants in this study has been published in the following article: Hastings, R. P., Totsika, V., Hayden, N. K., Murray, C. A., Jess, M., Langley, E., & Margetson, J. K. (2020). 1000 Families Study, a UK multiwave cohort investigating the well-being of families of children with intellectual disabilities: Cohort profile. *BMJ Open*, *10*(2), 1–10. <https://doi.org/10.1136/bmjopen-2019-032919>. This citation also provides full acknowledgements of those involved in data collection.

to challenge some of the assumptions made that these sibling relationships would be both worse as well as drastically different than other siblings' relationships.

Abstract

This study draws on data from the 1,000 Families Study examining the wellbeing of families of children with intellectual disability (ID). In the current study, we explore whether the behavioural and emotional problems and prosocial behaviours of child sibling pairs (N of pairs = 500), where one child has ID, are associated with their sibling relationship quality. The Strengths and Difficulties Questionnaire measured internalizing and externalizing problems, and prosocial behaviours for both children in sibling dyads. Sibling relationship quality was measured using dimensions of the Sibling Relationship Questionnaire (SRQ) focused on intimacy-companionship and antagonism-quarrelling. Family poverty, child gender, birth order (sibling older or younger than the child with ID), and disability type were also incorporated in the analyses as controls. Confirmatory Factor Analysis (CFA) indicated that the model fit for the latent variables measuring sibling relationships were adequate. The final structural model indicated that the prosocial behaviour and internalizing problems of the children with ID, their typically developing siblings' prosocial behaviours, and sibling birth order were associated with intimacy-companionship in the sibling relationship. The internalizing and externalizing problems, and prosocial behaviour of the children with ID, their siblings' externalizing problems, and sibling birth order were associated with antagonism-quarrelling in the sibling relationship. Longitudinal analyses are required to understand these associations further.

Introduction

Intellectual Disability (ID) is characterized by reduced intellectual function and reduced everyday adaptive behaviour with an onset before the individual's 18th birthday (AAIDD, 2019). A meta-analysis of international studies suggested that the prevalence rate for ID is approximately 1% (10.37/1000 population, 95% CI 9.55, 11.18 per 1000 population) of the global population (Maulik et al., 2011). USA-based population data (Platt et al., 2019) suggest this figure may be higher, estimating the prevalence rate is 3.2%. Individuals with ID may also have developmental disabilities (DD), such as autism, Down syndrome (where ID is always present), Cerebral palsy, Fragile X syndrome, or (for young children) global developmental delay.

Families of children with intellectual and developmental disabilities (IDD) have been the focus of academic study for several decades. Empirical studies indicate parents and siblings may be at risk of worse psychological outcomes than in families of children without ID (Singer & Floyd, 2006; Emerson et al., 2010; Hayden et al., 2019a). In terms of family roles, siblings are likely to become increasingly important in the lives of their brothers and sisters with ID. Improvements in health and life outcomes for disabled people (e.g. increased life expectancy amongst people with Down syndrome; Hodapp & Urbano, 2007), along with reduction in state welfare available in various national contexts, has prompted questions surrounding the uncertainty of care and support for people with DD (Power & Bartlett, 2019). Although the future role of siblings is uncertain, researchers have speculated that siblings may be increasingly called upon for support and care roles (Leane, 2019). Closer sibling relationships between siblings where one has an ID may increase the likelihood that the sibling without ID will be willing to take on this caring or supporting role in the future (Burke et al., 2012). Therefore, understanding and fostering sibling relationships in this group is vital from a policy and practice perspective.

Understanding sibling relationships is also practically important for both siblings, as well as being of theoretical interest. Sibling relationships are unique in that they are the only relationship that potentially lasts across the entire life-span, from early childhood into old-age. Siblings have the potential to influence one

another's lives (Meltzer & Kramer, 2016). Children learn and develop social behaviours within their sibling relationships (Mandleco & Mason Webb, 2015) that may influence their social behaviours and relationships beyond their immediate family and childhood. Furthermore, general sibling research indicates that sibling relationship quality is associated with mental health outcomes, such as depression – in childhood as well as adulthood – adjustment problems, internalizing and externalizing behaviours, and substance abuse (Feinberg et al., 2012; Waldinger et al., 2007). Sibling relationships may be particularly salient in families of individuals with ID. Richardson and Jordan (2017) emphasized the way in which sibling relationships may be 'imperative to the lives of people with disabilities' (2017: 1536). Beyond their potential to provide care, siblings have the potential to offer friendship and support where elsewhere in society, disabled people may face exclusion and discrimination.

The current research is informed by family systems perspectives. Family systems perspectives explore the way in which family members, and family sub-systems, may influence and relate to one another (White & Klein 2002; Smith-Acuña, 2011; Cox & Paley, 1997). An assumption underlying many ID family research studies, is that the primary direction of influence will be of the child with ID on their family, including their siblings, and that this will be a 'negative' effect (Hastings, 2016). There are data supporting a (relatively small) potential negative impact of living with a disabled brother or sister on siblings' well-being. Recent population based studies have explored the behavioural and emotional outcomes of siblings of children with ID (Hayden et al., 2019a) as well as the mental health outcomes of siblings of children with DD (Marquis et al., 2019) in comparison to siblings of children without disabilities. However, such research focused on individual family members does not fully reflect systems thinking (Rosenblatt, 1994) and a more complete understanding is needed through exploration of relationship outcomes, such as by exploring the sibling dyad. Systems thinking encourages us to conceptualize the sibling-dyad level relationship as reciprocal and interconnected in nature (Cox & Paley, 1997; Cox, 2010) and to move beyond focusing entirely on the effect of the disabled sibling on their brothers and sisters.

Existing quantitative research on sibling relationships often includes measures of relationship dimensions of warmth, closeness, conflict, status/power,

and rivalry (Furman & Buhrmester, 1985). These dimensions were developed distinctively by Furman and Buhrmester (1985) to avoid the ‘negative’/‘positive’ divide of relationship dimensions scales prior to this measure. For example, a sibling relationship, using their measure, can be understood as both highly warm and highly conflictual. Whereas prior relationship scales would measure warmth and conflict together to define a relationship as warm *or* conflictual. Therefore, this conceptualization allows various dimensions of the relationship to be understood, and it allows for ambivalence, as well as detachment and disinterest by either sibling in the sibling relationship.

In the non-disability literature, a meta-analysis found that where children’s and adolescents’ sibling relationship quality had more warmth and less conflict, this was associated with lower internalizing and externalizing problem scores for participants (Buist et al., 2013). Therefore, sibling relationships are an important predictor for understanding children’s and adolescents’ behavioural and emotional outcomes. Studies exploring sibling warmth and closeness when one has a disability have found particularly positive sibling relationships, for example, when one sibling has Down syndrome (Hodapp & Urbano, 2007) and this may be related to behaviours associated with Down syndrome, such as higher levels of prosocial behaviours. Although many researchers have hypothesized that sibling relationships where one child has an ID might be more ‘negative’, the empirical evidence base is currently unsubstantiated. Existing research exploring more ‘negative’ aspects of the sibling relationship have found similar levels of conflict between typically developing sibling pairs compared to sibling pairs where one has an ID (Doody et al., 2010). Other studies have found that sibling pairs where one has an ID may have less conflict in their sibling relationships than other siblings (Floyd et al., 2009; Kaminsky & Dewey, 2001).

An important question to consider is: What factors are associated with different dimensions of sibling relationship quality for siblings when one has an ID? Most of the research exploring the effect of the sibling relationship or disabled siblings’ outcomes have focused on how these factors relate to the non-disabled siblings’ psychological and social outcomes. For example, Hayden et al., (2019a) found that older siblings of children with ID had increased total behaviour problem scores, peer problems, and conduct problems compared to older siblings of children

without ID. These siblings' problem behaviour scores were associated with the elevated problem behaviour scores of their brothers and sisters whether or not they had ID. A smaller-scale study suggested that higher levels of behaviour problems in autistic children were associated with less warmth and closeness in the sibling relationship and more conflict in the sibling relationship (Hastings & Petalas, 2014), and behaviour problems in autistic children have also been associated with poorer sibling adjustment for the non-disabled sibling and poorer quality in the sibling relationship (Jones et al., 2019). There has been relatively fewer studies about the effect that the sibling relationship has on *both* siblings when one has an IDD. One rare study found that conflict in the sibling relationship was associated with internalizing behaviour problems in the non-disabled sibling and in externalizing behaviour problems in the sibling with ID (Begum & Blacher, 2011).

We identified only two studies that provided data about the effect of children's behaviours on their sibling relationship. Orsmond et al. (2009) found that sibling relationships were more positive where autistic siblings had fewer behaviour problems. We also identified a study that found that the behaviours of siblings of children with Williams syndrome were associated with their sibling relationship quality (Cebula et al., 2019). Cebula et al. (2019) found that siblings' self-reported prosocial behaviours were associated with more warmth in the sibling relationship, as reported by their parent/carer. They also found that siblings' self-reported total behavioural difficulties was associated with more self-reported conflict in the sibling relationship. Further work needs to be done to examine this effect using a broader and larger IDD sample. These studies therefore provide some evidence that the behaviors of both the child with IDD and their sibling will be associated with their sibling relationship quality. Family systems perspectives would suggest that both siblings' behavioural and emotional problems would have an association with their sibling relationship quality and that these effects would be reciprocal. To resist a negative narrative about the effect a person with IDD may have on their non-disabled brother or sister without IDD, analyses should examine effects that include both siblings' behavioural and emotional symptoms in more equal ways.

The primary aim of the current study was to explore whether, in sibling dyads where one has an ID, the behavioural adjustment of both siblings is associated with their sibling relationship quality. We expected that both siblings' behavioural

and emotional problems and/or prosocial behaviour would be associated with sibling relationship quality.

Method

Participants

Participants were 500 primary caregivers reporting on nearest-in-age sibling dyads where one has an ID, and both were aged between 4 years and 15 years and 11 months. Table 5.1 summarizes descriptive statistics for the sample included in the current study. The primary caregiver was usually the biological mother of the child with ID (primary caregiver is biological mother = 89.0%; primary caregiver female = 94.0%). The mean age of siblings was 9.23 years ($SD = 3.12$; range = 11.00) and the mean age for the children with ID was 8.66 years ($SD = 2.68$; range = 11.00). There were more males amongst the children with ID (66.8%) whereas 47.1% of siblings were male. Almost half of the children with ID had a severe/profound ID as reported by their primary carer (49.0%). In terms of additional diagnoses, 50.6% of children with ID were reported by their primary carer to have autism and 17.6% were reported to have Down syndrome. Sixty eight (13.6%) of the siblings were reported as having a longstanding illness, disability or infirmity. This included siblings with conditions such as dyslexia ($n = 6$), ADHD ($n = 10$), health conditions such as diabetes ($n = 1$) or asthma ($n = 5$), or mental health conditions such as anxiety ($n = 6$). In terms of the ethnicity of primary caregivers, 93.1% were White, 3.4% were Asian and 2.0% were Black.

Measures

Behavioural and emotional adjustment. Primary caregivers completed the Strengths and Difficulties Questionnaire (SDQ; parent version; Goodman, 1997) to measure the emotional and behavioural adjustment of both the children with ID and their nearest-in-age sibling. The SDQ is a 25-item screening questionnaire with items rated on a three-point scale: 'Not True', 'Somewhat True' or 'Certainly True'. Three SDQ scores were used: internalizing problems (emotional and peer problems), externalizing problems (conduct and hyperactivity problems), and prosocial behaviours. Items on the internalizing problems scale included 'Many worries' and 'Rather solitary, tends to play alone'. Example items from the externalizing problems scale included 'Often lies or cheats' and reverse-coded 'Thinks things out

before acting'. Items on the prosocial behaviour scale included 'Considerate of other people's feelings' and 'Shares readily with other children'. Goodman et al. (2010) argued internalizing/externalizing domains should be used in community samples and where users are not screening for disorder because these scores have more satisfactory convergent and discriminant validity than the individual SDQ subscales in community contexts. The SDQ has also been shown to have good validity for identifying behavioural and emotional problems in children with ID (Murray et al., 2020). In the current study, for the SDQ responses about the children with ID, the internal consistencies (Cronbach's α) for internalizing problems was .77, externalizing problems was .73, and prosocial behaviour was .85. For the SDQ responses about the siblings, the internal consistencies (Cronbach's α) for internalizing problems was .83, externalizing problems was .86, and prosocial behaviours was .85.

Sibling relationship quality. Items were selected from the Sibling Relationship Questionnaire (SRQ; short form; Furman and Buhrmester, 1985) to assess relationship quality in the sibling dyads. Although the short form SRQ has 39 items, 10 items were included in the survey to reduce participant burden. These 10 items have been used previously in an autism sibling study (Petalas et al., 2012). Two questions measured sibling intimacy: 'How much do the sibling and the child tell each other everything?' (SRQ1) and 'How much do the sibling and the child share secrets and private feelings?' (SRQ2). Sibling companionship was measured using two questions: 'How much do the sibling and the child go places and do things together?' (SRQ3) and 'Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do the sibling and the child play around and have fun with each other?' (SRQ4). Sibling affection was measured by asking: 'Some siblings care about each other a lot while other siblings don't care about each other that much. How much do the sibling and the child care about each other?' (SRQ5) and 'How much do the sibling and the child love each other?' (SRQ6). Sibling quarrelling was measured by asking 'How much do the sibling and the child disagree and quarrel with each other?' (SRQ7) and 'How much do the sibling and the child get mad and get in arguments with each other?' (SRQ8). Finally, sibling antagonism was captured by asking 'How much do the sibling and the child insult and call each other names?' (SRQ9) and 'How much

are the sibling and the child mean to each other?' (SRQ10). Primary caregivers respond to all statements about the sibling relationship on a five-point Likert-type scale: 'Hardly at all', 'Not too much', 'Somewhat', 'Very much' or 'Extremely much'. Internal consistency (Cronbach's α) in this sample was .84 for intimacy, companionship and affection in the sibling relationship, and .84 for quarrelling and antagonism in the sibling relationship.

Poverty. Poverty was measured by combining four distinct measures: income poverty; subjective poverty; ability to raise emergency funds and; deprivation (See Table 5.1; Emerson, 2003; Emerson, 2004; Hayden et al., 2019a). Income poverty estimated whether the family were living below or above the UK median weekly household income. Subjective poverty was measured by asking primary caregivers how well they were 'managing financially these days' on a five-point scale ('living comfortably', 'doing alright', 'just about getting by', 'finding it quite difficult', or 'finding it very difficult'). Responses were dichotomized, with those families responding that they were 'finding it quite difficult' or 'finding it very difficult' categorized as 'struggling financially' with all other responses categorized as 'managing financially'. Primary caregivers were asked if they could raise £2,000 in an emergency with four possible responses. These response were dichotomized with those that responded that they could 'easily raise the money', or could by making 'sacrifices' being grouped together as able to raise the money and those that selected 'I don't think I could raise the money' or could only do so by doing 'something drastic' coded as struggling to raise the money. Neighbourhood deprivation was measured using data from the index of multiple deprivation. This uses postal code area data to measure neighbourhood deprivation in small locales within each country in the UK. These data provide relative deprivation information based on seven indicators: income, employment, education, health, crime, housing, and environment (Gill, 2015). A dichotomous variable was derived with those in the bottom quintile (higher levels of neighbourhood deprivation) of neighbourhoods for the UK country in which they lived vs. the remainder (lower levels of neighbourhood deprivation). All four of these deprivation measures were then summed into one composite variable with values ranging from zero to four (higher numbers mean the family had more indicators of poverty).

Procedure and study design

Data for this study were from the first wave of the 1,000 Families Study, a UK-based, large-scale, ongoing longitudinal data set of families with at least one child with ID living full-time in the household (Hastings et al., 2020). The overall study sample size at Wave 1 included 1,184 primary caregivers. Once we removed those cases where the child with ID did not have a brother or sister, there were 612 sibling pairs. A further 99 cases were removed because the parent had indicated that the sibling themselves had an IDD. A further 13 cases were removed because they were missing measures needed for the analysis. The sample for the current study consisted of 500 sibling pairs where one had an ID and one did not.

The primary caregiver was the main study respondent and provided all study data. Study materials were in English, so primary caregivers needed to have suitable English language skills to participate. Primary caregivers also had to be aged 18 years and over to be included in the study. Families were recruited using a multi-point method: through websites and social media (i.e. Facebook and Twitter), via UK research and charity organizations' newsletters, and through special schools. Most caregivers completed the survey online, although to remove barriers to participation, primary caregivers could request postal surveys.

The study adhered to the ethical standards of the British Psychological Society. The UK National Health Service (NHS) West Midlands—South Birmingham Research Ethics Committee provided full ethical approval and we also received institutional approval prior to the commencement of data collection (see Appendix Four, 4.1-4.14 for study documentation and ethics). Primary caregivers provided fully informed consent before they took part in the study, and best-practice confidentiality and data protection standards were adhered to throughout.

Analysis procedure

Structural equation modelling (SEM) was employed. SEM benefits from being theoretically driven (Kline, 2016). Analyses were conducted using IBM SPSS AMOS 26 using maximum likelihood estimation, and a two-stage process was taken. First, Confirmatory Factor Analysis (CFA) was used to ascertain whether the SRQ items sufficiently measured the latent constructs related to sibling relationship quality. Further adjustments to the measurement model were required to improve model fit and the loading of items in relation to the latent constructs. Fit was

measured using statistics available from our models where means and intercepts were estimated to account for missing data, and included the Tucker Lewis Index (TLI; $\geq .95$), the Comparative Fit Index (CFI; $\geq .95$), and the Root Mean Square Error of Approximation (RMSEA; $\leq .06$). These cut-off levels indicated were recommended by Hu and Bentler (1999), and were subsequently endorsed by Cabrera-Nguyen (2010).

Once the latent constructs measuring sibling relationship quality were identified and tested, we progressed to the second analysis stage where structural models were fitted to examine the main research question exploring associations between the behaviours of the siblings and the children with ID with their sibling relationship quality. Variables were included in the models iteratively using a forward entry approach. First, we incorporated the SDQ internalizing, externalizing, and prosocial behaviour scores – for the children with ID and their siblings with the latent constructs encapsulating sibling relationship quality. The second structural model included control variables in one model to identify which variables were significantly associated with the latent constructs measuring sibling relationship quality. The control variables measured were informed by existing research and were: sibling birth order (Braconnier et al., 2018), the children with ID and their siblings' gender (Floyd et al., 2016; Orsmond & Mailick Seltzer, 2000; Cuskelly & Gunn, 2003), family poverty represented by combining four distinct measures: income poverty; subjective poverty; ability to raise emergency funds and; neighbourhood deprivation (See Table 5.1; Emerson, 2003; Emerson, 2004; Hayden et al., 2019a), and whether the children with ID also had Down syndrome, or autism (Orsmond & Seltzer, 2007; Hodapp & Urbano, 2007). The final structural model incorporated each of the control variables and behaviour variables that met the threshold of $p < .10$ in terms of their association with the sibling relationship latent constructs.

Results

Measurement models

Figure 5.1 illustrates the initial measurement model for the 10 items from the SRQ as two latent constructs (intimacy, affection and companionship; and antagonism-quarrelling). The model fit was not sufficient ($\chi^2(34) = 834.46$; $p < .001$;

CFI = .74; TLI = .58; RMSEA = .22). The factor loadings ranged from .42 to .99, indicating some of the items were not adequately associated with the latent constructs.

Alternative models and associations were explored to improve the model fit of the measurement model including a unidimensional model, removing items with low factor loadings, and aggregating items (i.e., parcelling; Matsunaga, 2008). Parcelling has the benefit of improving model fit, stabilizing parameter estimates as well as offering potential psychometric benefits (Matsunaga, 2008).

The final measurement model is shown in Figure 5.2. For the latent construct measuring intimacy, affection and companionship, the items measuring affection – items five and six – were removed from the measurement model as they had the lowest factor loadings (.51 and .47 in Figure 5.1 respectively). For the latent construct measuring antagonism-quarrelling in the sibling relationship, parcelling was used to improve the model fit of the construct. Both the ninth and tenth items were capturing ‘antagonism’ in the sibling relationship, these two items were parcelled to better distil the latent construct (Matsunaga, 2008). These changes improved the model fit ($\chi^2 (13) = 97.40; p < .001; CFI = .96; TLI = .91; RMSEA = .11$) indicating the items were adequately measuring the latent constructs.

Structural models

The structural models exploring predictors of sibling relationship quality are summarized in Table 5.2. The first stage involved identifying the SDQ scores of the child with ID and their sibling that were ($p < .10$) associated with the latent constructs measuring sibling relationship quality (see Table 5.2, Models 1.0 and 1.1.). This stage confirmed that the child with ID’s prosocial behaviour and internalizing problems and the sibling’s prosocial behaviour were associated with the intimacy-companionship latent construct. Model 1.1 ($\chi^2 (32) = 83.72; p < .001; CFI = .98; TLI = .96; RMSEA = .06$) also confirmed that the child with ID’s prosocial behaviour, and externalizing and internalizing problems, and the sibling’s externalizing problems, were associated with the latent construct encapsulating antagonism-quarrelling in the sibling relationship. Some pathways were, therefore, removed from the variables that remained in the final structural models (e.g. the pathway between sibling externalizing problems to intimacy-companionship was removed) and one

variable was completely removed from the final structural models (i.e. sibling's internalizing problems). All the retained pathways were statistically significant ($p < .001$).

The second stage involved identifying the control variables that would be retained in the final structural models by removing those with associations with a p value $>.10$. Table 5.2 provides a summary of this stage (models 2.0 and 2.1). Model 2.1 ($\chi^2(37) = 154.57; p < .001$; CFI = .94; TLI = .90; RMSEA = .08) confirmed that the following variables were to be retained in the final structural models: the child with ID having autism ($p = .036$), sibling gender ($p = .055$), and sibling birth order ($p = .012$) were associated with the latent construct measuring intimacy-companionship in the sibling relationship; and family poverty ($p = .006$) and sibling birth order ($p < .001$) were associated with the latent construct measuring antagonism-quarrelling in the sibling relationship. Other control variables were not retained in the final structural models.

The final structural models are summarized in Table 5.2 (models 3.0 and 3.1). Further control variables were removed after model 3.0 ($\chi^2(65) = 221.49; p < .001$; CFI = .95; TLI = .89; RMSEA = .07) where the p value for some of the associations was $>.05$. This resulted in family poverty, sibling gender, and autism being removed from the final structural model.

The model fit for the final structural model (model 3.1 in Table 5.2) was adequate ($\chi^2(47) = 199.00; p < .001$; CFI = .95; TLI = .89; RMSEA = .08). Figure 5.3 provides a visual summary of the final structural model, as well as standardized estimates of the associations between the variables and latent constructs measuring sibling relationship quality. Table 5.3 provides these standardized estimates and the p values for these associations.

The child with ID's prosocial behaviour had the strongest positive association with intimacy-companionship in the sibling relationship ($\beta = .49, p < .001$). The sibling's prosocial behaviour ($\beta = .21, p < .001$) and the child with ID's internalizing problems ($\beta = .11, p = .011$) were also positively associated with the latent construct measuring intimacy-companionship in the sibling relationship. The child with ID's externalizing problems had the strongest association with antagonism-quarrelling in the sibling relationship ($\beta = .36, p < .001$), and their prosocial behaviour ($\beta = .31, p <$

.001) and internalizing problems ($\beta = .19, p < .001$) were also significant paths in the model, along with the siblings' externalizing problems ($\beta = .17, p < .001$).

For sibling birth order, where the sibling was younger than their brother or sister with ID, this was associated with both more intimacy-companionship ($\beta = .11, p = .013$) and more antagonism-quarrelling ($\beta = .13, p = .001$) in the sibling relationship.

Discussion

In the present study we explored whether the behavioural and emotional adjustment of children with ID and their closest-in-age sibling was associated with their sibling relationship quality. The final structural model (Figure 5.3) showed that intimacy-companionship in the sibling relationship was higher where the child with ID had higher levels of internalizing problems, and when both the child with ID and their sibling had more prosocial behaviours. Antagonism-quarrelling in the sibling relationship was higher when the child with ID had more internalizing problems and more prosocial behaviours, and when both the sibling and their brother or sister with ID had higher levels of externalizing problems. For each of these pathways in Figure 5.3, the associations were positive (more problem or prosocial behaviour resulted in higher levels of the sibling relationship quality latent constructs). Thus, the behaviour of both the children in the sibling dyad was associated with sibling relationship quality. The birth order covariate indicated that where the sibling was younger, this was associated with both more antagonism-quarrelling and also intimacy-companionship in the sibling relationship.

The prosocial behaviour associations with antagonism-quarrelling may appear counterintuitive. However, it is feasible that the children with ID having higher prosocial behaviours may be associated with milder ID. Children with ID with higher social abilities may be able to engage with their siblings in a more equal way – and *vice versa* – with the sibling treating their brothers/sisters with ID more equally – including with antagonism-quarrelling.

The only covariate that remained in the final structural model was sibling birth order. This may be related to the way in which younger siblings may be more likely to spend time with, and treat their brother or sister with ID more equally, both arguing and playing together more freely. Older siblings may take on more caring or responsibility-based roles, whereby they disengage or avoid antagonism-quarrelling

with their brother or sister with ID. The other covariates were not statistically significantly associated with sibling relationship quality.

In terms of theoretical implications, these findings support a family systems perspective whereby both siblings' behaviours (both the more 'negative' and the more 'positive' aspects) have an association with both 'positive' and 'negative' aspects of the sibling relationship. Although family systems perspectives can be used to model reciprocal associations about sibling relationships when one has an IDD such as in this study, it is interesting that family systems perspectives are rarely applied in this way, but rather in an arguably over-simplified way that assumes a negative narrative. It may be valuable to consider the non-disability sibling literature, which favours social learning theory (Feinberg et al., 2012) over family systems perspectives to theorize and model the way siblings' relationships with one another are associated with both siblings' behaviours. Social learning theory assumes that both siblings learn and affect one another in an equal way, emphasizing reciprocity. It may be worth considering why this theory is applied commonly in the general sibling literature, but not generally in the IDD and disability sibling literature. One reason may be that sibling IDD researchers have assumed a negative, one-sided, and non-reciprocal narrative about siblings of children with IDD. Reciprocity is the essential premise of social learning theory, whereas for family systems perspectives, it is one component of an overall concept. Future research should seek to ensure that family systems perspectives are not used simply to justify a focus on siblings' outcomes but, rather, are fully engaged with in a meaningful way to inform research design, analyses, and interpretations of findings, ensuring that reciprocal effects at the sibling-dyad level are further understood.

Limitations.

A limitation of the current study is that the models were from cross-sectional data. Longitudinal data would be required to provide further support to the final model and importantly, to ascertain the direction of the pathways beyond the essentially theoretical model provided by these cross-sectional data. The final structural model presented is also only indicative at this stage due to its 'adequate' model fit. Therefore, other variables or model configurations may explain and fit the data better.

The data included in this study were from primary caregiver report only. We know that parents and children report on children's sibling relationships differently (Rankin et al., 2017; Tsai et al., 2016; Cebula et al., 2019). There are of course pragmatic reasons (e.g. ease of administration, access and ethics, research costs) why parental report was the most suitable solution to collect large-scale quantitative data, such as that in this current study. In the current design, children's behaviour is being interpreted by their primary caregiver. For example, a child sharing a toy with their brother or sister may score highly in prosocial behaviours as perceived by a parent, but if the reason for their sharing is to avoid arguments, they may be indicative of avoidant behaviours (perhaps associated with internalizing problems), that are not being identified as such by their primary caregiver. A further example is the way in which relationship indicators seen as 'negative', such as quarrelling or antagonism, may be helpful ways for children to communicate their needs and frustrations or to learn from siblings about how to manage human relationships more generally (this possibility may also support the suggestion that future sibling IDD research could be considered using social learning theory). A parent on the outside of this relationship would inevitably perceive the relationship differently than siblings themselves.

The measurement model for sibling relationship quality required items to be parcelled and dropped to improve item loadings onto the latent constructs and to improve the model fit of the latent constructs. Even after improvements, the model fit indicated the model was adequate. Therefore, we question whether the SRQ is the most appropriate measure of sibling relationship quality for this sample. Measuring something as ambivalent and dynamic as sibling relationship quality, particularly from the perspective of someone outside of that relationship (i.e. primary caregiver reported data) is challenging enough before we begin to also consider the associated measurement challenges when one of those children in the sibling dyad has an ID. For example, items surrounding quarrelling may have been difficult for some primary caregivers to respond to if their child with ID was minimally verbal. The model fit also has conceptual problems as well. We were essentially left with a construct that measured 'positive' and 'negative' dimensions of sibling relationship quality, and so the measurement of sibling relationship in the current study lacks nuance.

A further limitation of the current study is the focus on dyadic sibling relationships. Family systems theorists would argue that more ‘triadic and whole family interactions’ (Cox & Paley, 1997: 249) need to be studied to fully understand the family system, and the sibling relationship in turn.

Future research directions.

The limitations outlined provide direction for further research. For example, longitudinal data would be required to further support the model presented in this study to help us understand further the direction of the pathways. Longitudinal data would also allow an exploration of how sibling relationships change over time. Future research should consider if there are more effective ways of measuring sibling relationships where one sibling has an ID. This could involve providing further guidance to primary caregivers about how to interpret the items on the SRQ where one child has an ID, or this may require a new questionnaire to be developed specifically for sibling relationships where one sibling has an ID. Sommantico et al. (2020) developed the Siblings’ Experience Quality Scale specifically for non-disabled siblings of adults with IDD, chronic physical illness, or mental illness to examine siblings’ relationships. Using CFA, they demonstrated the measure’s effectiveness when tested on a mixed sample of adult siblings of people with IDD, chronic physical illness, or a mental illness. However, this measure has been designed for and validated with an adult sample.

In future, researchers should also find ways to incorporate self-report responses, not just from the non-disabled sibling, but also from their brother or sister with ID. A child self-report questionnaire may inadvertently risk excluding the perspectives of the children with ID due to the reading and comprehension level required to complete a questionnaire without support. This may reinforce the exclusion of disabled people in research and wider society (Meltzer & Kramer, 2016; Richardson & Jordan, 2017). Future qualitative research would be an important way of developing methods to incorporate both siblings’ perspectives on the sibling relationship, with the flexibility to be appropriate for the differences and preferences of both the sibling and their brother/sister with ID. Finally, future studies may explore models that incorporate functioning across multiple family members and family sub-systems, as well as models conceptualized at the sibling-dyad level.

Applying theoretical frameworks such as family systems perspectives and social learning theory may inform complex, reciprocal, dyad-level studies.

Practical and clinical implications

Any intervention developed to improve relationship quality at the level of the sibling dyad ought to be cognizant of and involve the full family system. This approach is supported by data exploring associations between sibling behaviours and relationships with parenting practices, and sibling relationships with marital relationships (*cf.* Feinberg et al., 2012; McHale et al., 2012). The final structural model in this study provides some information that may be used to develop interventions to support sibling relationship quality when one sibling has an ID. For example, prosocial behaviour from both siblings was associated with sibling relationship quality, suggesting an intervention that focused on increasing prosocial behaviour in both the child with ID and sibling may be fruitful. The importance of birth order in the final structural model also supports a differential approach depending on whether the sibling is older or younger than their brother or sister with ID. For example, where the sibling is older than their brother or sister, interventions may explore facilitating more contact between the siblings, whereas when the sibling is younger than their brother or sister with ID support may take the form of facilitating conflict management in the sibling dyad.

Tables and figures

Table 5.1. Sample descriptive statistics

Demographic and family descriptive statistics	
Mean age of sibling in years (<i>SD</i>)	9.23 (3.12)
Mean age of child with ID in years (<i>SD</i>)	8.66 (2.68)
Sibling is older	56.7%
Sibling is male	47.0%
Sibling has a longstanding illness, disability or infirmity	13.6%
Child with ID is male	67.1%
Child with ID has severe/profound ID	49.0%
Child with ID has autism	50.6%
Child with ID has Down syndrome	17.6%
Primary caregiver is female	94.0%
Primary caregiver is the biological mother	89.0%
Family has no indicators of poverty on the poverty composite. Poverty indicators:	26.2%
Family would struggle to raise £2,000 in an emergency	48.4%
Family indicated that they were struggling financially	11.5%
Lowest quintile for neighbourhood deprivation	14.7%
Household is below the median for weekly household income	62.5%
Primary caregiver is White	93.1%
Primary caregiver is Asian	3.4%
Primary caregiver is Black	2.0%

Table 5.2. Structural models predicting sibling relationship quality

Structural models	χ^2 (<i>df</i>)	Model <i>p</i>	CFI	TLI	RMSEA	All paths <i>p</i> <.10
1.0 All behaviours of child with ID and sibling.	86.12 (33)	<.001	.98	.95	.06	No
1.1 Behaviours of child with ID (int., ext., pro.); sibling (ext., pro.)	83.72 (32)	<.001	.98	.96	.06	Yes
2.0 All control variables.	163.57 (49)	<.001	.95	.89	.07	No
2.1 Control variables: sibling birth order, sibling gender, autism, poverty.	154.57 (37)	<.001	.94	.90	.08	Yes
3.0 Significantly associated behaviours of child with ID, sibling and control variables (see variables included in models 1.1 and 2.1).	221.49 (65)	<.001	.95	.89	.07	No
3.1 Final model (Figure 5.2): sibling: int., pro., child with ID: ext., int., pro., and sibling birth order.	199.00 (47)	<.001	.95	.89	.08	Yes <i>p</i> <.05

Note. All behaviours included: Int. = SDQ internalizing behaviours. Ext. = SDQ externalizing behaviours. Pro. = SDQ prosocial behaviours. All control variables included: Single or two parent household, poverty composite, sibling birth order, child with ID gender, sibling gender, whether the child with ID had autism, whether the child with ID had Down syndrome.

Table 5.3. Final structural model associations between predictors and sibling relationship quality latent constructs

Predictors of sibling relationship quality	Association with intimacy- companionship in the sibling relationship Standardised regression weights (<i>p value</i>)	Association with antagonism- quarrelling in the sibling relationship Standardised regression weights (<i>p value</i>)
Sib Externalizing behaviours	-	.17 (<.001)
Sib Prosocial behaviours	.21 (<.001)	-
Child with ID Internalizing behaviours	.11 (.011)	.19 (<.001)
Child with ID Externalizing behaviours	-	.36 (<.001)
Child with ID Prosocial behaviours	.49 (<.001)	.31 (<.001)
Sib Birth Order	.11 (.013)	.13 (.001)

Figure 5.1. Initial measurement model, latent constructs measuring sibling relationship quality

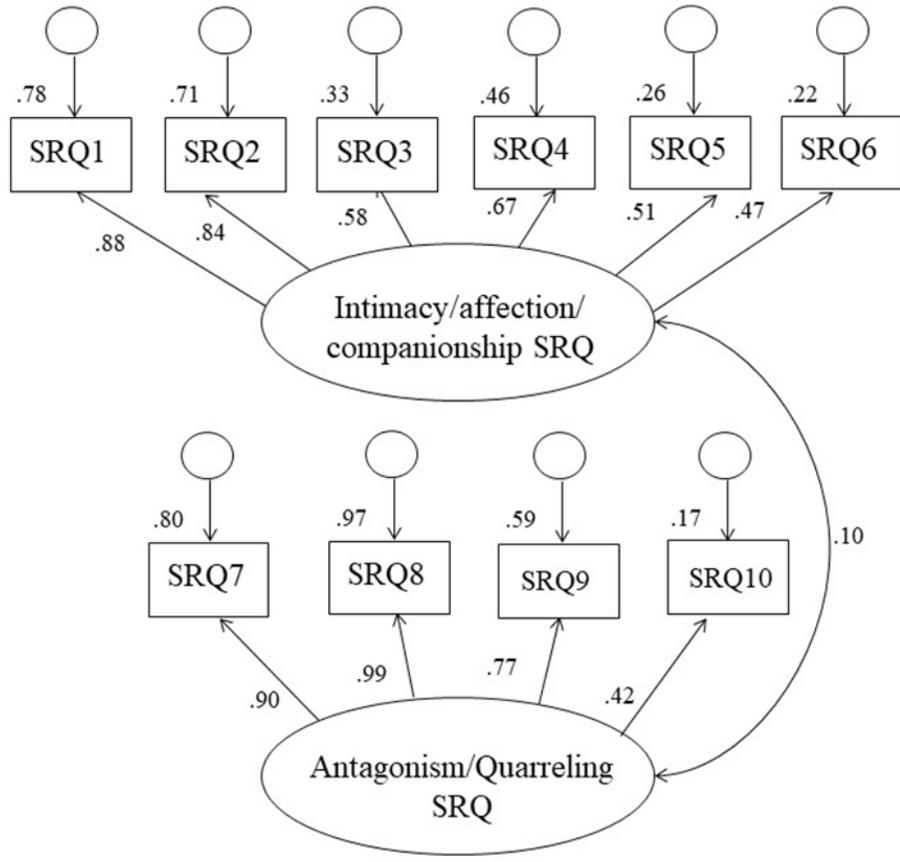


Figure 5.2. Final measurement model, latent constructs measuring sibling relationship quality

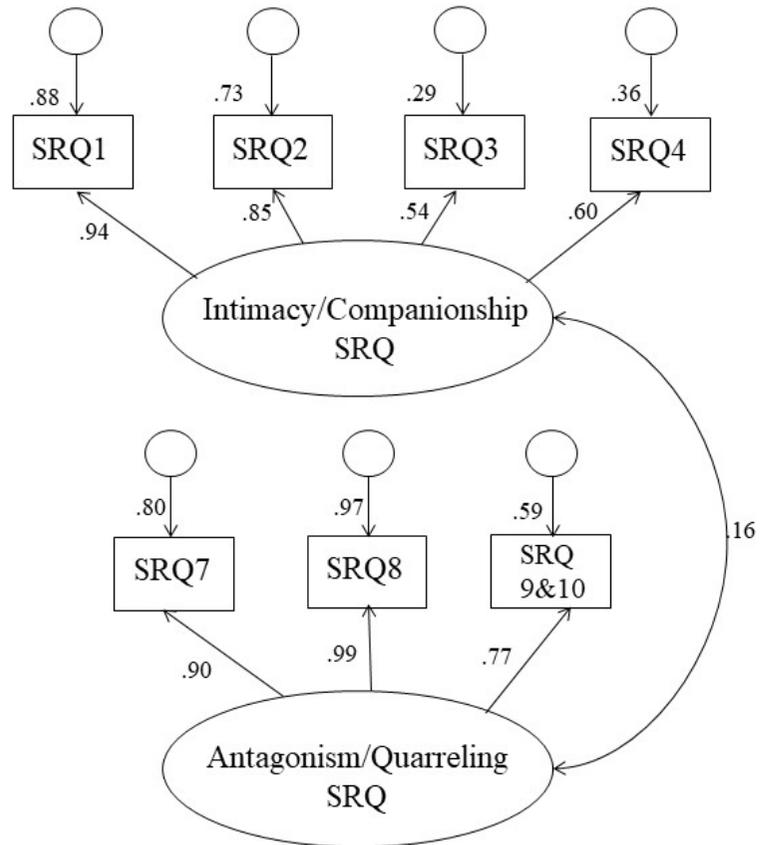
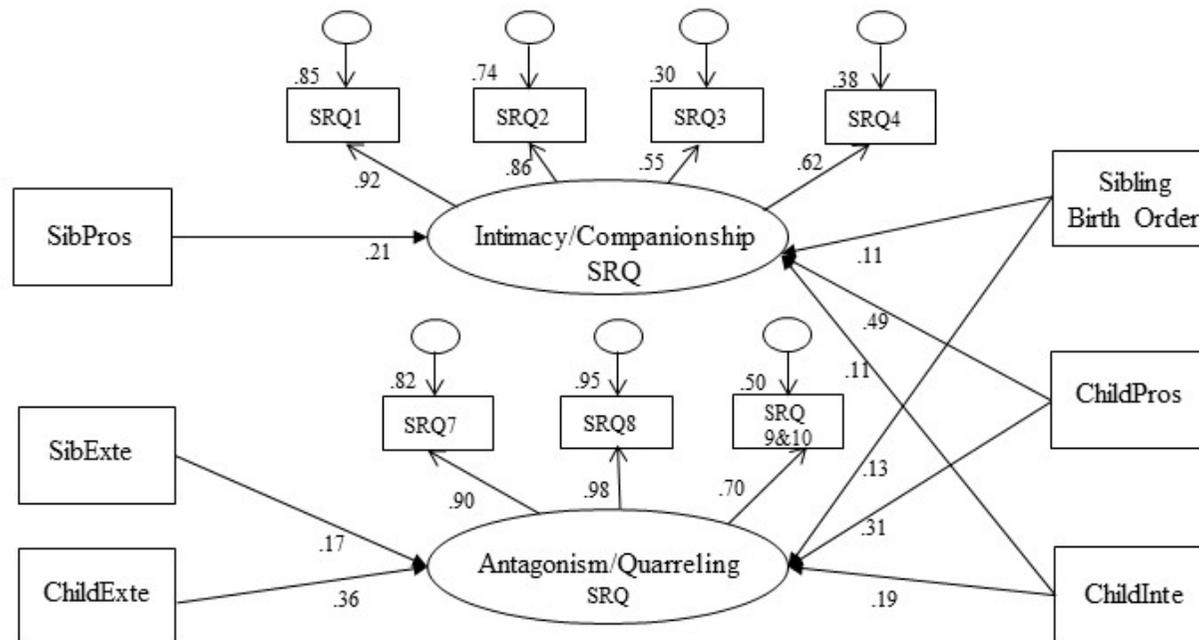


Figure 5.3. Final structural model: Associations between the behaviours of sibling pairs and their sibling relationship quality where one has ID.



Note. SibPros = Sibling prosocial behaviours SDQ score; SibExte = Sibling externalizing behaviours SDQ score; ChildPros = Child with ID prosocial behaviours SDQ score; Child Exte = Child with ID externalizing behaviours SDQ score; ChildInte = Child with ID internalizing behaviours SDQ score.

Chapter 6: Discussion

Cumulatively, this thesis provides new data about the psychological, health and social outcomes of siblings of both children and adults with IDD. The first chapter provided an overview of three areas that have guided the development of this thesis: theoretical perspectives, existing empirical literature, and my own experiences as a sibling of an autistic person. Chapters two to five included four distinct empirical studies exploring the outcomes of siblings of people with IDD. These studies included: a large-scale population study providing the first population-level data about older siblings of children with ID; an evaluation of a small-scale one-to-one school-based intervention, embedded in a wider discussion about school-based experiences for siblings; a large-scale survey study about the mental distress, wellbeing, quality of life, and health outcomes of adult siblings of people with IDD using moderated regression analyses; and lastly, a more theoretically-driven analysis using large-scale survey data to explore the reciprocal associations between both siblings' behavioural and emotional outcomes on the sibling relationship, using structural equation modelling. This final chapter seeks to draw together findings and issues in these prior chapters and explore its cumulative contribution, theoretically, practically, and empirically. Whilst considering the limitations of these studies, this chapter will also consider what is next for sibling IDD research, before returning to personal reflections about being a sibling of an autistic person.

Overview of Thesis Chapters and Contribution

Chapter one provided an exploration of key ideas, knowledge, and experience that have informed and that contextualise the work in this thesis. It began by providing an overview of existing theories most commonly applied to family research: family systems perspectives, the Double ABCX model, as well as lifespan and developmental frameworks. These theories enabled a conceptual understanding of why siblings may be affected by having a brother or sister with IDD, as well as emphasised the importance of sibling relationships from a theoretical perspective. These theories also supported an approach that explored sibling outcomes beyond childhood, and provided guidance about which factors to include in statistical analyses in order to better understand siblings' outcomes. Conceptual issues, such as the foundational tendency for sibling research to rely on a negative narrative, were also discussed. Chapter one then provided an overview of existing empirical studies relevant to the focus of the overall thesis. This overview of the existing literature was

by no means exhaustive, it focused primarily on reporting on methodologically robust studies, such as population-based data, large-scale data and systematic reviews. This review highlighted the need for further population-based data analyses and large-scale data to answer questions about siblings, as these types of data have reduced biases and more statistical power, and allow for more advanced analysis techniques to be employed.

Cumulatively, the theoretical discussion and empirical discussion highlighted the need to explore more complex and interesting research questions, such as indirect associations using moderation analyses or exploring reciprocal effects using structural equation modelling. The third section of the introduction chapter explored my personal experiences as a sibling of my autistic brother. I attempted to provide an open, self-reflexive examination of my experiences with a view of understanding my own perspectives and assumptions about my research further. This section also highlighted some methodological issues related to the need for quantitative social sciences research to acknowledge and utilise traditionally qualitative epistemological understandings of the knowledge our work produces. This piece of self-reflexive writing could be understood in relation to the theoretical frameworks discussed as well as the existing empirical research discussed. Quantitative research could do more, however, to explore some of the nuances related to my own experiences, such as by exploring reciprocal effects and considering the family within an Embedded Systems Framework (Kovshoff et al., 2017).

The second chapter presented findings from the first of four empirical studies. This study used data from the Millennium Cohort Study, a nationally representative data set, to understand whether older siblings of children with ID had worse outcomes than other children (Hayden et al., 2019a). This study is essentially premised on testing the assumption about whether or not the non-disabled sibling is negatively affected because they have a brother or sister with ID. Although there are a plethora of studies that test this, the existing research is inconclusive, with findings being mixed, likely due to methodological biases. The main strength of our study therefore, was that it was the first study published internationally, to ask these questions using large-scale, nationally representative data, reducing issues with sampling biases. We found that older siblings of children with ID had slightly worse outcomes for some, although not all, behavioural and emotional symptoms compared

to older siblings of children without ID. However, when we accounted for other factors that might be related to psychological difficulties, the siblings of children with ID had similar levels of difficulties as other siblings.

This population-based study benefited from having a range of covariates included in the analyses that were driven by the theoretical perspectives discussed in chapter one, such as maternal mental distress, single parent households, as well as a well-defined measure of socio-economic position. Overall, our findings suggest that siblings of children with ID were only slightly more at risk of psychological difficulties than other siblings. By using clinical cut-off scores as well as reporting mean scores for siblings' behavioural and emotional symptoms, we were able to identify that most siblings of children with ID were scoring within the 'normal' range. However, a small number of older siblings of children with ID might benefit from some help and support, as this group were 1.5-2 times more likely to be scoring in the 'abnormal' range for some of the behavioural and emotional symptoms. Very importantly, it seemed that what might explain sibling difficulties where they did exist were factors related to having a brother or sister with ID (e.g., more likely to live in a poorer household) rather than simply the presence of having a brother or sister with ID. We did not find a simple story of increased problems only because a sibling has a brother or sister with an ID. More recently, other population-level data about siblings of children with DD have also been published (Marquis et al., 2019; Marquis et al., 2020). These studies benefit from drawing on mental health information as measured by health services rather than self-report, they had a sample of both younger and older siblings, and they were able to explore some questions about the differences in sibling outcomes based on the specific DD that their brother or sister had.

Chapter three contained the second empirical study, an evaluation of *Sibs Talk*, a one-to-one, school-based intervention for siblings of children with disabilities. This evaluation is embedded within a wider discussion about siblings' school experiences and sibling supports. Given our findings in the first empirical study in this thesis (chapter 2), there are a small group of siblings in need of additional support. This is the first intervention, to our knowledge, that is one-to-one, allowing for a relationship between the sibling and a school staff member to develop, rather than being family-based or group-based. This may provide specific benefits,

such as allowing for some continuation of support through these relationships with school staff members to continue beyond the length of the intervention. The second unique aspect of this intervention, is that it was school-based, and to our knowledge, this was the first school-based intervention for siblings of children with disabilities. Given the school-based needs siblings may have (Goudie et al., 2013; Gregory et al., 2020) as well as the conceptualisation of schools being a part of siblings' mesosystem (Kovshoff et al., 2017) the school context may provide a useful setting to provide siblings with support.

The results presented in chapter 3 indicate improvements in the siblings' SDQ and HIFAMS scores from the point in which they began the Sibs Talk intervention to the point in time they had the final intervention session. Four of the eight measures showed improvements for siblings from the beginning of the intervention to the end of the intervention: emotional symptoms; hyperactivity; prosocial behaviours and; SDQ total behavioural and emotional difficulties. The improvements seen were small to moderate in terms of effect size. Written comments were also collected from the participating siblings. Overall, the data presented in this evaluation indicate that Sibs Talk may have contributed to positive outcomes for children who took part in the intervention. These comments also provided some further context about siblings' experiences and their relationship with their brother or sister with a disability. Although challenges were mentioned by siblings, particularly when they experienced physical aggression from their brother or sister with a disability, many positive comments were also made by siblings about their brother or sister with a disability, particularly in relation to their relationship with their brother or sister with a disability.

The third empirical chapter (chapter four) is the only empirical study about adult siblings in this thesis. Chapter one discussed lifespan and developmental perspectives which supported the inclusion of both adult and child studies in this thesis. The data for this study is about 911 adult siblings that took part in an online survey. To our knowledge, this is the largest sample of UK adult siblings, providing more power in our analysis models and allowing us to include a large number of covariates in our analyses, as well as interaction terms. A strength of the study was that, as with chapter three, this study was co-produced with the UK charity Sibs, with further feedback on the research design provided by additional adult siblings

that were not involved with the design of the study. As we used existing measures of mental distress, wellbeing, quality of life, and health, we were able to compare our findings to population norms without having to recruit a sample of other adults. Using these measures allowed us to provide context to siblings' average scores, reporting simple differences in mean scores between siblings and other adults. This may seem like an obvious finding to want to report, but most of the adult sibling studies already in existence did not provide these comparisons as they did not use existing, validated measures, nor did they recruit normative comparison or control groups.

We also disaggregated and reported carers' and non-carers' outcomes as well, because the existing empirical literature does not provide a clear understanding on how sibling carers' outcomes compare to siblings that are not carers. It is sometimes important to ask very simple questions of our data in order not to assume that siblings have worse outcomes across the outcome variables. For example, although siblings had worse outcomes on the mental distress and wellbeing outcomes, their health and quality of life was broadly similar to those adults in the 'normative' samples. This sort of data is also most useful for charities and organizations such as Sibs, as they can more easily communicate siblings' needs to funders, as well as make decisions about which aspects of support they ought to focus on, allowing further impact work to stem from this research.

The research questions we asked were slightly more developed than in the studies contained in chapters two and three. We used moderated regression to examine whether SES and the level of independence of the brother or sister with IDD moderated the association between carer status and siblings' outcomes. This allowed us to go beyond direct effects and consider which groups of siblings may be in need of additional supports, without assuming that all siblings will be in need of supports. After all, our findings in chapter two did not support this assumption. As in chapter two, chapter four also found that SES was an important variable in understanding siblings' outcomes. Again, a strength of this study is that we incorporated various indicators of SES and had a developed definition, relative to other IDD sibling studies, of SES that moved beyond income. Another strength, was that we used an established definition of carer status, from the NHS, rather than allowing siblings to self-define what caring meant to them, a problem in some of the

existing sibling carer literature (Lee et al., 2019b; Lee & Burke, 2018). As with chapters two and three, chapter four considered psychological factors as the main outcomes.

The final empirical study, chapter five, moved beyond examining psychological outcomes for the non-disabled sibling, focusing on sibling relationships as the main study outcome. The way this analysis was conceptualized was the most theoretically-driven and went the furthest to avoid making assumptions about sibling dyads where one sibling has an ID. There are relatively fewer empirical studies about sibling relationships in the IDD family research field compared to studies concentrating on the psychological outcomes of non-disabled siblings. As discussed in the literature review in chapter one, we found no empirical evidence to suggest that sibling relationships where one sibling has an IDD were any worse than sibling relationships where neither sibling was disabled. The model hypothesized in chapter five resisted a simple negative narrative (Hastings, 2016) that siblings were negatively affected simply because they have a brother or sister with IDD. Instead, we hypothesized that both siblings' 'positive' and 'negative' behaviours may be associated with both 'positive' and 'negative' aspects of their shared sibling relationship. This mirrors the way sibling relationships have been conceptualized in the general, non-IDD sibling literature, that sibling relationships are ambivalent and reciprocal. Our analyses supported such a conceptualization. This is important, because it indicates that it is not simply the sibling with ID having a negative effect on their non-disabled sibling, but that both siblings are having a reciprocal effect on the sibling dyad in both 'positive' and 'negative' ways, supporting a family systems perspective. Although chapter two included the behavioural and emotional problems of the brother or sister with IDD as a covariate, chapter five is the only chapter that included data about both siblings in a more equal way. Sibling studies that include data about both the sibling with ID and the non-disabled sibling equally, such as in this study, remain uncommon.

Taken together, these studies contributed to the knowledge base about the psychological outcomes of child and adult siblings. For child siblings, we also explored some social outcomes, in the form of their sibling relationships, for adult siblings, we also examined quality of life and health outcomes. A strength of this thesis has been its reliance on large-scale datasets to explore siblings' outcomes

(chapters two, four, and five), including one nationally representative population data set (chapter two). For chapters two, four, and five, we were also able to examine socio-economic status in a more developed way than existing sibling studies have been able to do. As indicated in chapters two and four, this is an important variable in understanding the experiences of siblings of people with IDD, and as we will discuss, is an area for future research in order to understand how to better support siblings of people with IDD. Importantly, these studies do not support a negative narrative that all, or most, siblings will experience worse outcomes simply because they have a brother or sister with IDD. Although some siblings are in need of extra support, suggestions about which will be made in the practical implications section below, most siblings are physically and mentally well. The findings of this thesis did not find support for a simple negative narrative about siblings of people with IDD.

Theoretical Implications

Each of the empirical chapters within this thesis drew on family systems perspectives, albeit this is reflected on somewhat perfunctorily in the writing for the first three empirical chapters. This is conventional in family and sibling IDD research, which has been critiqued as ‘atheoretical’ (McHale et al., 2016: 589; Saxena & Adamsons, 2013: 300). This section will extend these theoretical discussions about the empirical chapters, applying the theoretical concepts and conceptual issues presented in chapter one.

The first empirical chapter, chapter two, applied logistic regression to examine whether older siblings of children with ID experienced worse behavioural and emotional outcomes than siblings of children without ID. The study was positioned using family systems perspectives to justify our research questions focusing on siblings and our hypothesis that they may experience worse outcomes than other children because of the presence of a brother or sister with ID. This premise falls utterly under the negative narrative of which I have attempted to resist. However, there was value in asking these, somewhat problematic, questions again – as this study was the first to ask these research questions using a robust, nationally representative dataset. It is important to ascertain if this group of children are at risk and in need of supports. It was also important to consider what other factors may explain the group differences we found.

Family systems perspectives informed the covariates we selected to consider whether the group differences we found would remain once we controlled for other factors. For example, within-family factors, such as whether the sibling and child lived in a single parent home, the mental distress of their mother, and the behavioural and emotional problems of the child with or without ID, were included in the analyses. The Embedded Systems Framework (Kovshoff et al., 2017) supported the inclusion of within-sibling factors, such as their gender, and social factors in the macrosystem, such as wealth and social class. The stress theories we examined in chapter one, specifically the Double ABCX model, also supported exploring family resources (bB) and the pile up of stressors (aA) that may exist if families of children with ID are also experiencing material deprivation, or a parent is experiencing mental distress. In this way, our analyses being informed by theoretical approaches was an important factor in resisting a negative narrative, as we found that once we adjusted our analyses for these covariates, the effects of the older sibling having a brother or sister with ID or not were non-significant in five out of six models. This provided further support for applying theoretically driven modelling in future analyses, and also to consider indirect effects where appropriate data is available to do so. Specifically, the findings from chapter two reinforced the importance of applying multi-layered systems thinking to studies about siblings.

My second empirical chapter (chapter three) discussed school-based experiences for siblings and evaluated Sibs' school-based, one-to-one, intervention 'Sibs Talk'. The intervention and evaluation could also be understood using a multi-layered systems perspective. If we draw upon the Embedded Systems Framework again (Kovshoff et al., 2017), the intervention addresses within-sibling factors, such as how siblings interpret events, and their psychological wellbeing. It considers factors related to siblings' microsystem, by considering siblings' home lives with their family, and their school lives with their teachers and peers. It also considers the siblings' mesosystems, i.e., how these areas interact. Again, the premise of this intervention could be justified using a family systems perspective, suggesting that siblings may be in need of extra supports due to having a brother or sister with a disability. It could also be understood using the Double ABCX model, whereby Sibs Talk becomes a resource that siblings can apply to the situation (bB), it could help siblings interpret their situation (cC) and could help siblings adapt to their situation

(xX). Our findings from this study, where we saw small to medium effect size improvements in the emotional and behavioural outcomes of the siblings that took part in the intervention, provided support for considering the school context as a site for intervention in the future, which is consistent with a multi-layered systems approach to sibling research.

The third empirical chapter (chapter four) is the only adult sibling study within this thesis. Aside from the obvious statement that all adults were once children, there are theoretical reasons for including an adult sibling study in a thesis predominately formed of child sibling studies. Developmental and lifespan perspectives consider the ways in which families change over time, including changing family positions, roles, and norms. We may expect to see over the life course, that the role non-disabled siblings take in the lives of their brothers and sisters with IDD will change over time. We may expect that many young adult siblings move out of the family home, and have a more independent life whilst their parents are still able to care for their brothers and sisters with IDD. Over time, and as parents age, siblings may increasingly take over aspects of care for their brothers and sisters with IDD. Although many child siblings provide informal care for their brothers and sisters with IDD, adult siblings are more likely to take on a more formal care role than child siblings.

For the adult sibling survey study presented in chapter four, we can also apply the systems approaches and stress models I have already discussed. Again, our first research question was similar to that asked in our first empirical chapter, which was whether adult siblings were at risk of worse outcomes compared to other adults. Again, we used a family systems perspective to inform this approach. We found that, particularly for the mental distress and wellbeing outcomes, the siblings did have worse outcomes (medium to large effect sizes) compared to other adults. Again we justified asking psychological research questions about siblings despite the risk of advancing a negative narrative because we had relatively strong data to explore these questions. Although this sample was not nationally representative, to our knowledge, it is currently the largest UK study sample ($N = 911$) about adults of people with IDD, and it is the first study we have identified to make meaningful comparisons to normative samples about the outcomes of adult siblings of people with IDD.

Our covariates were selected and our moderation model was designed using systems perspectives, although the Double ABCX model, again, could be used to explain our findings. For example, the Double ABCX model would suggest that being a carer (aA), the person being cared for having lower levels of independence (aA), and experiencing lower levels of socio-economic deprivation (aA, bB) could potentially lead to worse psychological and health outcomes because there is a 'pile up' of demands or stressors. Again, multi-level family systems perspectives can also be applied to help inform why we selected the test and covariate variables that we did. For example, we included within-sibling variables, such as gender and level of education, family-based variables, such as the sibling and their brother or sister with IDD being the same or a different gender, as well as wider social factors, such as socio-economic status, and neighbourhood deprivation. We can also use family systems perspectives and stress models to explain our findings. For example, we found that for siblings experiencing the highest levels of socio-economic deprivation, that being a carer was no longer associated with the siblings' outcomes. This may indicate that the stress and hardships related to socio-economic deprivation are so great, that the effects of caring were no longer statistically significant. In terms of disrupting a negative narrative about the effects of having a brother or sister with IDD, this is important. Both this study, and our first empirical study (chapter two) indicated that social, structural factors were accounting for more of the variance than any of the within-person variables of their brother or sister with IDD e.g. autism, Down syndrome, level of independence. This study supports the application of theoretical concepts to consider indirect factors, such as moderating effects as in this empirical chapter, when exploring the outcomes of siblings of people with IDD.

My final empirical study, chapter five, had the most theoretically driven analyses. Again, for this analysis, family systems perspectives were applied to design the conceptual model whereby the behavioural and emotional outcomes of both the child with ID and their brother or sister without an IDD were modelled to be associated with both the 'positive' and 'negative' aspects of their sibling relationship. In this way, their sibling relationship has been conceptualised in a more similar way to the more general sibling literature, rather than the IDD sibling literature. This is because the relationship is the focus and outcome rather than the psychological outcomes; there is assumed reciprocity, and there is assumed

ambivalence and equality in the sibling subsystem. Examining conceptualisations of sibling relationships in this way is important for disrupting a negative narrative about families where one member has an ID, as research focusing on the siblings' psychological outcomes risks positioning the child with ID as a 'problem' and the non-disabled sibling as a 'victim' of the relationship. Family systems perspectives inform a focus at the sibling dyad level, and also encourage us to consider the bidirectional nature of the sibling relationship, and encourage analyses to explore reciprocal effects, as we have done in this empirical study. So often in the IDD sibling literature, however, this reciprocal nature of the sibling dyad is overlooked.

Again, we drew on systems thinking to identify covariates to include in our model. It was interesting that only sibling birth order met our statistical inclusion criteria to remain in the model, because this is a dyad-level variable that both siblings are equally a part of. Covariates about the sibling with ID, such as having Down syndrome or autism, did not sufficiently contribute to the analyses in order to remain in the final structural model. We also included socio-economic status as a covariate, which can be justified by both systems thinking and stress model thinking, and it was also not sufficiently associated to be retained in the final structural model. This is interesting as it tells us that structural factors are not necessarily associated with siblings' relationships, although chapters two and four indicate this is an important factor in understanding siblings' behavioural and psychological outcomes. Overall, the findings of the final empirical study support further systems based thinking which emphasises the reciprocal nature of the sibling relationship.

Cumulatively, the theoretical implications of the empirical studies within this thesis indicate that systems, stress, developmental, and lifespan perspectives can enable us to conceptualise our studies and interventions as well as help us understand and contextualise our findings. Stress theories such as the Double ABCX model help us to consider why family resources and socio-economic factors were such important variables in chapters two and four. Caution should be taken not to use stress theories to apply a pathological, deterministic framework to sibling research. Developmental and lifespan perspectives help us to consider why we should focus on adult (chapter four) and child siblings (chapters two, three, and five). Although we often included as a covariate the age of the sibling to understand how siblings' outcomes may vary

based on age, future research should consider siblings' experiences across stages of the lifespan.

The theories we used most consistently and explicitly throughout the thesis were family systems perspectives. As in most sibling IDD studies, it was used to inform a focus on siblings' psychological outcomes for chapters two, three, and four. A multi-level, embedded (Kovshoff et al., 2017) systems perspective was used to inform a focus on school based factors in chapter three. Family systems perspectives can also be used to focus on sibling relationships (chapter five), as exploring experiences at the sub-system level, and considering reciprocal effects in that sibling dyad are considered valuable in systems-level theories, because only by understanding parts of the family system can the family be understood as a whole (Smith-Acuña, 2011; White & Klein, 2002). As with the Double ABCX model, family systems perspectives helped us to identify covariates and helped us to explain our findings.

There is a risk, however, that I have at times fallen into throughout this thesis – as all sibling IDD research does at times – and that is in assuming a negative narrative about the effect that having a brother or sister with IDD can have on a sibling. Family systems perspectives can be used to advance this negative narrative if it is applied too simply, however, it also has the capacity to disrupt a negative narrative, particularly when it is used to inform more advanced statistical analyses that are capable of examining more interesting research questions, such as those exploring indirect effects, and those exploring reciprocal effects. Chapter five highlighted the importance of considering reciprocal effects in the IDD sibling research field. This aspect of reciprocity in family systems theory must not be lost when conceptualising siblings and families of children and adults with IDD.

Practical Implications

A strength of this PhD has been that it was collaborative with the UK charity Sibs. This not only means that they have been involved in the research design and data collection of chapters three and four, but also that the findings of this thesis may have meaningful impact through the work of this charitable organisation. The findings from chapters two and four indicated that some siblings may be at risk of worse outcomes compared to other people. The question therefore becomes: what

sort of supports or intervention should we put into place for these siblings? As our findings indicate that most siblings are not experiencing worse psychological outcomes compared to other children and adults, a blanket offer of clinical support would not be justified and would be overly deterministic. Despite the focus on psychological factors related to siblings of people with IDD throughout this thesis, there is not necessarily sufficient support for clinical interventions for siblings specifically. Although, of course, siblings that are also experiencing mental health problems should be able to access appropriate psychological interventions whether or not these problems are associated with their experiences as a sibling.

A scattergun approach to supporting siblings is not only impractical in a context where social and charitable funding is limited, but may not be desired, as we would want to avoid an unsubstantiated negative narrative that all siblings need intervention. A focused approach to sibling supports may be more appropriate. The important covariates in chapters two and four may provide guidance on where organisations may focus their support. Poverty and SES were associated with psychological outcomes for participants in both chapters two and four. Therefore, organisations may decide to focus their support on siblings that are also from low SES households, or organisations like Sibs may consider developing specific projects to support siblings that are experiencing low SES and poverty. For chapter two, maternal mental distress and the sibling being from a single parent household were also important covariates in understanding why some siblings had worse psychological outcomes compared to other siblings. This supports an embedded systems perspective being taken into account during intervention design. Rather than focusing on the sibling as an individual, potentially a family-systems based intervention would be appropriate, whereby the whole family are involved in an intervention. Chapter five indicated that there was an association between both the sibling with ID and the non-disabled siblings' psychological outcomes and their sibling relationship quality. This indicates that it is important to include the disabled sibling in an intervention and consider fostering family relationships, and not merely focusing attention on individual family members' psychological outcomes.

The findings from chapter three indicated that Sibs Talk may be one potential source of support for child siblings. There are pragmatic reasons why we would not expect the uptake of Sibs Talk to be high. For example, the one-to-one aspect of the

intervention within the context of austerity, and now a global pandemic, means fewer schools will be able to offer staff-time for ten sessions of one-to-one support. Online supports, such as groups, events, and peer mentoring, may be particularly relevant, and practically in the current global context, it may be the only feasible option for supporting siblings from a community perspective at this time. Some families living in poverty may not have regular, private access to online supports, and so these types of support may exclude the most vulnerable siblings. Potentially, part of sibling support may involve campaigning at a political and structural level to reduce poverty and enhance state welfare for all people. It is justifiable for sibling support to include working to enhance the lives of disabled people, given that population-level data indicates families of people with ID experience more socioeconomic deprivation (Emerson, 2003; Emerson, 2004) and from a family systems perspective, the outcomes and experiences of disabled people affect their families, including siblings. Organisations such as Sibs may decide to dedicate some resources to collaborate with other disability-related organisations to campaign at a more structural and political level to enhance the lives of disabled people and their families as well as to continue supporting siblings in a practical and focused way.

Realistically, however, limitations in funding and resources for siblings will result in siblings continuing to be supported in an *ad hoc* way. This should involve a multi-level and co-ordinated approach to sibling support. Interventions may involve focused support for siblings most at risk, for example, those living in lower SES households (chapters two and four). For child siblings, those living in single parent households and those where a parent is experiencing mental distress may also be the focus of intervention (chapter two). Considering the impact of these broader family factors in siblings' outcomes, an intervention may include a whole family intervention that draws upon family systems perspectives to work with the whole family. Chapter five may provide support for an intervention that focuses at the sibling dyad level to foster closeness in sibling dyads where the older sibling is non-disabled and to reduce conflict in sibling relationships for sibling dyads where the younger sibling is non-disabled. Interventions should be more focused and consider family and wider contextual factors related to siblings' experiences and outcomes.

Overall, clinical and pathological approaches may not be appropriate interventions for supporting siblings generally, and focusing on 'fixing' siblings,

when most siblings are doing well, is not appropriate. This is because it is highly individualistic and ignores some of the structural reasons why some siblings may need support, such as indirect factors related to socio-economic disadvantage. Furthermore, advancing a negative narrative, a pathological narrative, or an overly deterministic narrative about the outcomes of siblings, and seeing them as individuals that need ‘fixing’ stigmatises disability and mental health problems. Some siblings will experience psychological problems and receive clinical diagnoses for mental health problems, and these siblings are likely to receive psychological and/or psychiatric support through medication and/or talking therapies. Therefore, clinical support ought to be more needs-led for siblings, as their needs for support may not be directly related to having a brother or sister with an IDD.

Limitations and Directions for Future Research

Each of the empirical chapters described the specific limitations of each study as well as provided suggestions for future research relevant to the research questions posed in each study. This section seeks to emphasise the overarching limitations, as well as discuss implications for research, making suggestions for future research.

Analysis Considerations

All of the survey studies within this thesis drew upon cross-sectional data. Using longitudinal analyses in future research would enable us to conduct more advanced analyses exploring, for example, mediating effects. Therefore, longitudinal data (i.e. three or more time points) should be collected to further our understanding about how siblings’ experiences change over time. Longitudinal data would provide further support for our models. For example, in chapter five, we used structural equation modelling to theorise that siblings’ behavioural and emotional outcomes would be associated with their sibling relationship quality. Longitudinal data would be required for us to confirm whether the effect is in the direction that we hypothesised, or whether another configuration would explain the data better. For chapter three, where we presented a pre-post evaluation design about Sibs Talk, although the study showed small to medium sized positive effects for siblings that took part, we do not know whether the effects would be maintained over time. Longitudinal data would be required to understand these effects further. Although we

did explore reciprocal effects using structural equation modelling and we undertook moderated regression analyses, there are other advanced statistical analyses that would allow us to deepen our understanding of siblings, such as mediation analyses. Future research about siblings may be further influenced by family systems thinking and include analyses at the family level, rather than the sibling outcome level (chapters two, three, and four) or the sibling dyad level (chapter five), drawing on multi-level modelling approaches.

Sampling

Although we were able to draw on large-scale data for three out of four of the studies (chapters two, four, and five), for chapters four and five, these data were drawn from surveys drawing on a convenience-based sample of people. The biggest limitation of survey data primarily recruited online, is that they are more affected by sampling biases. Only one of our studies drew on nationally representative data (chapter two) and chapter three remained small-scale. Where possible, large-scale, nationally representative data should be drawn upon to understand siblings of people with IDD. Nationally representative datasets are limited, however, in that their existence is finite and that the measures are pre-defined for a broader purpose, and therefore, there is a need to still draw upon non-representative large-scale survey data in order to be able to examine more nuanced research questions relevant to families of people with IDD. The question then becomes: how do we ensure that these samples are more representative?

Sibling IDD research, and indeed social research more generally, consistently fails to recruit participants from marginalized communities. As researchers, we must find ways of effectively and meaningfully working with people that we have, inadvertently, excluded from our research. There have been only a handful of studies that have provided data specifically about Black and Asian siblings in the USA (Sonik et al., 2016, Richardson & Stoneman, 2019, Sage & Jegatheesan, 2010, Jegatheesan & Witz, 2013), and I have identified no comparable studies focusing on the experiences of siblings from ethnic minority groups in the UK. There have been cultural comparative studies, such as comparing the experiences and psychosocial adjustment of siblings of children with autism in the UK and Taiwan (Tsai et al., 2016; Tsai et al., 2018) and studies about siblings living in the global South e.g.

India (Dickinson, 2020). Studies such as these are important for understanding how sibling experiences and outcomes may differ based on socio-cultural factors. Future research should seek to both make general sibling samples more representative in terms of ethnicity and religion, but also specific studies should be conducted to understand the experiences of Black, Asian, and where geographically applicable, Indigenous siblings' experiences in Western contexts, as well as the experiences of siblings in non-Western countries as well.

Another group of people under-represented in sibling research are people from lower SES backgrounds or those living in poverty. This group may be particularly difficult to retain, as has been identified in the Millennium Cohort Study (Hansen, 2012). This may cause the representativeness of, for example, future waves of the 1,000 Families Study, to decline. We encountered this difficulty of under-recruiting those from lower SES backgrounds in the adult sibling survey, where our sample were disproportionately middle class, and our highest attrition rates were for the questions about socio-economic position. In response to this, we added further guidance about why we were asking questions about socio-economic position, and reminded participants that they could skip these questions. We hoped that this would improve retention of participants, however, these changes led to a statistically significant increase in attrition in the survey, as 89.3% of participants completed the income question before the additional wording, compared to 79.6% of participants that completed the income question after the additional wording ($\chi^2 (1, N = 911) = 15.13, p < .001$). There is therefore, no easy fix to this problem.

The preceding point highlights various issues related to how we define SES and poverty, how we encourage participants from lower SES households to take part in research, and how we help all participants to engage with these questions and improve retention after we ask personal questions such as these. We know that it is especially important to understand this group of siblings, as the findings in this thesis indicate that lower SES is associated with worse psychological outcomes for both child and adult siblings (chapters two and four). Broader population-based data tells us that having lower SES and experiencing deprivation is a consistent and considerable risk factor for worse outcomes, such as being more likely to have mental health disorders (Skapinakis et al., 2006), health conditions such as type 2 diabetes (Connolly et al., 2000) as well as engaging with behavioural risk factors

related to poorer health, such as smoking more and eating fewer fruits and vegetables (Lakshman et al., 2011). We need to do further work to ensure the representativeness of our quantitative samples, but there is also scope to undertake qualitative research to understand the outcomes of siblings from lower SES families. Another solution to the problem about not recruiting representative samples, for both SES and ethnicity, may be to use weighting methods in future analyses. Although we were able to use the Millennium Cohort Study weights for chapter two of this thesis, no weighting methods were developed for the survey data analyses in chapters four and five.

Research Approaches: Qualitative, Quantitative, and Mixed Methods.

This thesis has almost entirely drawn on quantitative data, but there are important areas of research that should involve qualitative approaches. Firstly, as discussed, we need to understand the experiences of siblings from various backgrounds, including from various class, cultural, geographic, ethnic and religious backgrounds. There have been specific qualitative studies about sisters of people with IDD (e.g. Richardson & Stoneman, 2019; Pavlopoulou & Dimitriou, 2019; Pavlopoulou & Dimitriou, 2020; Chase & McGill, 2019, Connell et al., 2016, Cridland et al., 2016), but I have not identified any studies focusing on brothers of people with IDD specifically. There is a trend in adult sibling research to disproportionately recruit non-disabled sisters rather than non-disabled brothers. This may be related to sisters possibly having more regular involvement in the lives of their brothers and sisters with IDD (Lee & Burke, 2018; Hodapp et al., 2010) or perhaps being more likely to be a caregiver (Sonik et al., 2016; Burke et al., 2012). Research should seek to understand the experiences and outcomes of brothers in research.

Another group of siblings that we should consider developing specific research about are bereaved siblings. Sibs and I were contacted by several bereaved siblings whilst recruiting for the adult sibling survey in chapter four. I have been unable to identify specific support for bereaved siblings of people with IDD, nor research about this group. There is research, however, about bereaved parents of people with IDD (Reilly et al., 2008). This discussion has relevance in family systems theory too. For Rosenblatt (1994), being 'in' or 'out' of a family system is based on a continuum rather than a dichotomy. Therefore, if a sibling were to pass

away, they would still be in the family system and affecting the family system, even if they were no longer actively present in it. Qualitative research can provide insightful data about siblings' reflections about their lives and their experiences. Furthermore qualitative research has pragmatic value. Where populations are small or where researchers find it challenging to recruit participants from certain populations, small-scale qualitative research may be the only first-step researchers can take to understand a population better. Where little data exists, small-scale qualitative data about specific groups of siblings can provide scoping evidence to design further research and also gain an understanding of how to better involve people from excluded populations in our research.

Sibling relationships where one has an ID have been relatively less studied compared to non-disabled siblings' psychological outcomes, and this problem is reflected in my thesis. Only chapter five explored sibling relationships using a parent-report, quantitative sibling relationship measure (Furman & Buhrmester, 1985). There is also scope to conduct qualitative research in the future about sibling relationships. Much of the existing qualitative research exploring sibling relationships where one has an IDD have been criticized as there is a tendency for existing studies to concentrate only on the typically developing sibling. This reinforces the exclusion of disabled people in research and wider society, as well as limiting the research findings, only capturing one available perspective on the sibling relationship (Meltzer and Kramer, 2016; Richardson and Jordan, 2017). Where research does qualitatively research both siblings, there is a tendency for the sample to only include siblings where one has more moderate forms of IDD, or the methodologies observe rather than interview the siblings with ID. Previous research therefore lack the 'voice' of *both* siblings when one has a more severe ID. The tendency for researchers to involve only those with milder intellectual and developmental disabilities in qualitative research is due to methodological challenges related to differences in communication and understanding. Potential participants with more severe ID may also be non-verbal or may not understand interviewers' questions (Petalas et al., 2015), particularly those related to more abstract and conceptual aspects of their sibling relationship. Therefore to include people with ID, qualitative interviewing techniques would need to be re-conceptualized and adapted. Richardson and Jordan (2017) suggest that there are not only complexities resulting

from verbal, cognitive and emotional abilities, but also additional ethical challenges regarding informed consent, especially for those participants with ID (*cf.* Cameron and Murphy, 2007). One example study that did include adult siblings where one had more severe ID used a photo elicitation, observation and craft-activity methodology, rather than an interview that gained the perspectives of the sibling with severe ID directly (Tozer et al., 2013). Future research about sibling relationships should incorporate both siblings' perspectives and methodologies need to be extended to facilitate this.

Although this thesis has focused entirely on quantitative methods, with the exceptions of the content analysis conducted as part of chapter three, I acknowledge the value of qualitative approaches, particularly when conducting research about a group of siblings that we know little about as a research community, or for pragmatic reasons related to ensuring social justice and inclusion, such as with including the perspectives of the person with IDD in sibling relationships research. Even in this PhD, which is almost entirely quantitative, I had to draw on qualitative methods to meaningfully engage with participatory involvement through the Sibling Research Advisory Group and when conducting Vineland Adaptive Behaviour Scales interviews to collect data for the 1,000 Families study. It is important to be able to move between quantitative and qualitative methods where appropriate. The third paradigm sibling researchers should engage further with is mixed methods research. There has been little 'real' mixed methods research about siblings of children with IDD. By 'real' I mean whereby the quantitative and qualitative data are meaningfully integrated (Creswell et al., 2003; O'Cathain et al., 2010; Fetters et al., 2013; Bryman, 2006) and speak to one another, as 'where methods have been integrated, the whole can be greater than the sum of the parts' (Barbour, 1999: 40). Primarily, researchers should take an open and flexible approach to designing and conducting research using the best possible methods available to them to answer the specific research questions at hand, rather than being overly tied or committed to a specific discipline or preferred methodological approach.

Measures, Who Reports, and Scope

In this thesis, we explored sibling relationship quality quantitatively (chapter five). However, the confirmatory factor analysis conducted for this construct

required adaptation and was merely ‘adequate’ in terms of model fit. This may have been because we used the measure on a sample of siblings where one had an ID, and the measure was developed for sibling relationships where neither has a disability. There is a need to develop and validate measures about sibling relationship quality specifically about relationships where one sibling has an IDD. These need to include ways for the disabled sibling to self-report. These are not new points, both of these points were made more than a decade ago (Seltzer et al., 2005). Qualitative methods may provide more flexible approaches to allow the inclusion of the perspectives of people with IDD in research about their sibling relationships. Quantitative methods and survey questions that are able to be administered online in order to recruit relatively large samples do also have value in enabling us to understand sibling relationships further when one has an IDD. This does present issues however. For example, people with mild ID may need support to take part in online surveys, and people with moderate, severe or profound ID may not be able to take part in a survey at all. It is also difficult to gain parental consent in order to allow children to take part in online surveys, and they may also need extra support to participate in these questionnaires.

Sommantico et al. (2020) have developed a questionnaire for non-disabled adult siblings to report on their sibling relationship and experiences where they have a brother or sister with IDD, chronic physical illness, or a mental illness. They performed a confirmatory factor analysis to test the sibling relationship measure on a group of adult siblings of people with various disabilities, including IDD, and it had an excellent model fit. Further work needs to be done to validate a translated version of this measure from Italian to English. A questionnaire could also be developed that would be suitable to be administered to both members of sibling dyads where one sibling has developmental disabilities without ID or with mild ID. Extra support would need to be offered, and this may be a barrier to this questionnaire being used on large-scale studies. Another questionnaire about sibling relationships that would be valuable would be a sibling relationship questionnaire similar to the one developed by Furman & Buhrmester (1985) whereby a parent completes it about both siblings’ relationship with one another. Feedback from families that took part in the 1,000 Families study indicated that they found the Furman & Buhrmester (1985) questionnaire difficult to complete if, for example, their child with ID was minimally

verbal. Therefore a new questionnaire would need to be carefully developed in order to better apply to siblings where one has an IDD. This evidently creates problems related to neither sibling self-reporting, as well as it being odd to gather perspectives about a relationship from someone (i.e. a parent) that is not a part of that relationship. Despite these conceptual and pragmatic issues related to developing sibling relationship measures, I believe that they would still have value and improve the current sibling research about sibling relationships where one has an IDD and improve the experience for participants taking part in this type of research.

For pragmatic reasons, we have utilised parent-report data about child siblings (chapter two and five). Only chapter three provided some child-sibling self-reported data, along with teacher-reported data. Where possible in studies about child siblings, self-report data should be employed alongside parent-report and teacher-report data, as studies indicate that siblings often report differently than their parents (Rankin et al., 2017; Tsai et al., 2016; Cebula et al., 2019). Chapter four was about adult siblings and so that included sibling self-report, however, they also reported on their brother or sister with IDD. As highlighted when discussing sibling relationships, we must find ways of supporting siblings with IDD to self-report their experiences of their sibling relationships rather than relying entirely on the non-disabled siblings' perspectives on their sibling relationships (Meltzer & Kramer, 2016; Richardson & Jordan, 2017).

The range of measures used in this thesis was relatively restricted. For example, we used a measure of behavioural and emotional adjustment, the SDQ, throughout the child studies (chapters two, three, and five) rather than exploring various measures of adjustment or clinical diagnoses of mental health conditions. Qualitative written comments provided in the Sibs Talk study (chapter three) suggest that it may have been useful to measure additional areas beyond adjustment, such as knowledge about and attitudes towards, disability.

The scope of this thesis is, as all PhDs must be, limited and confined to a specific area. There are countless topic areas, measures and methodologies I could have employed, that are beyond the scope of this thesis. My thesis is, as the general sibling IDD research field is, saturated with studies about the psychological outcomes of child siblings. There were some emerging topics, such as about school

based experiences (chapter three), adult siblings' outcomes (chapter four), and sibling relationships (chapter five) that are relatively less explored in the sibling IDD literature. These are areas that more work needs to be done to further understand siblings' experiences.

Conceptual Issues

There are fundamental, problematic, conceptual issues with the way the focus on the study of siblings has been set up in every sibling study I have read, including in each of the preceding five chapters. Firstly, if we consider sibling studies from the perspective of the individual with IDD, it rests on a negative assumption (Hastings, 2016) that having a brother or sister with IDD may be a risk factor for worse outcomes for family members. There is also an argument to be made that it positions people with IDD as dependent on their siblings for support and care, and this fails to acknowledge the reciprocal potential of siblings' relationships. Sibling research has been considered as 'perhaps being in itself disablist in its orientation' (Sanders, 2004: 127) as it can be seen as marginalizing the disabled sibling whilst centring the non-disabled sibling. Sanders (2004) suggests to understand the effect on a non-disabled sibling, you would not just need to compare dyads of sibling pairs where one has a disability, with sibling pairs where neither has a disability, but also compare sibling dyads where both are disabled. Therefore analyses should be conducted on sibling dyads where both have an IDD as well as those where only one has an IDD.

For siblings, a focus on psychological outcomes rests on siblings being 'healthy' and best able to provide care for their brothers and sisters with IDD, which emphasises the ideological context much of the sibling research is situated in. By this, I mean the way in which families, rather than communities and societies, are expected to support disabled family members that verges on individualistic (rather than societal). We see this in the way that sibling studies are positioned, including my own. In order to get published, most sibling studies begin, not only by hypothesizing a negative narrative about the effect a disabled sibling may have on their non-disabled sibling, but by explaining that people with IDD are expected to outlive their parents, and siblings may then become carers for their brothers and sisters with IDD. In this way, an interest in siblings is justified by some contribution

siblings may provide which saves societal economic resources i.e. siblings' unpaid, informal labour.

These arguments are both gendered and racialized, given that population data tells us that carers are more likely to be women and Black (Sonik et al., 2016). Therefore, there is scope for more sociological and psychosocial theories to be employed to critique and understand sibling and family experiences further. Future research should consider how class, feminist, disability, post-colonial, critical race, and post-structuralist theories may be applied to sibling and family research in order to further understand siblings' experiences and disturb the assumptions we make about the families of people with IDD. There have been some important studies drawing on transformative and emancipatory perspectives (Richardson & Jordan, 2017) and criticisms of individualization and sibling disability work (Meltzer & Kramer, 2016). There is scope for further ideological critique about how siblings and families are expected to provide care.

Some of the theories I have used in setting up this thesis, such as family systems theory and the Double ABCX model, can fall into an individualistic narrative. I am using the concept of individualism as an antithesis to anti-community or anti-society discourse and policies, not its literal conceptualization of a single person being self-sufficient. Therefore, I see a focus on the immediate family (as opposed to government and community support) as the main source of support for a disabled person as individualistic. For example, the C in the Double ABCX model is about how families interpret their reality. Families can be seen to 'fail' to be resilient. It places the responsibility on the family, when arguably, the root problem is social if we consider our society from Marxist perspectives and from the perspective of the social model of disability, as unequal, disablist, capitalistic, and that undervalues disabled people and their carers. Future research should consider the potential of sociological theories to explore sibling IDD research in more critical ways and to resist negative narratives that may be inadvertently disablist, or by positioning siblings as only valuable as potential sources of free labour.

There are more pragmatic issues related to the conceptualisation of siblings as carers. In chapter four, our research questions explored sibling carer status and its association with sibling mental distress, wellbeing, quality of life, and health.

Although we used a clearly defined conceptualisation of carer status, using an NHS definition, we included a binary definition. Lee et al., (Lee et al., 2018; Lee et al., 2019b) argues that carer status should be understood as continuum rather than as a binary status. Carer status is not static either, the amount and type of role siblings play in their disabled siblings' lives varies throughout the lifespan. Future conceptualisations of caring in sibling IDD literature should consider the non-static nature of sibling carer roles.

As mentioned, in my own conceptualisations of each preceding chapter, I have arguably fallen into a negative narrative, whereby I justify a focus on siblings because of the potential deficit they face and because of the support and care they can offer their brother or sister with IDD. Although I believe that family systems theory can provide a way of avoiding a negative narrative about the effect that a person with IDD may have on their family members, a negative narrative can be intersected with family systems theory as well, as I focused on the psychological outcomes of the non-disabled sibling for much of this thesis. This is a problem that I cannot overcome, and no sibling or family researcher can overcome. However, family systems perspectives and family stress theories highlight that, only by ensuring the wellbeing of the whole family can individual members of it be supported and well, including the disabled family member. Therefore, I have had to take a pragmatic approach to these conceptual cracks of this thesis. People with IDD matter and have needs, as do their brothers and sisters, and, as highlighted by the findings in chapter five, they are interconnected. To ignore research about siblings because of its shaky, problematic, foundations, would not only do a disservice to non-disabled siblings, but also disabled siblings, and the family systems that they are a part of. Applying more critical, sociological, and psychosocial theories to what remains, a relatively 'atheoretical' (McHale et al., 2016: 589; Saxena & Adamsons, 2013: 300) research area, is an important next step to understanding and acknowledging some of the problems related to family IDD research.

Personal Reflections

I came to studying this PhD having studied law and then an education degree in a psychosocial studies department that was highly sociological, theoretical, and political. When I began this PhD I was new to statistics and psychology, felt more

committed to qualitative methods, and I was sceptical of post-positivist approaches to research, which I felt that IDD family research was embedded in. Learning about statistics having initially learnt about qualitative methods and epistemologies means that I am acutely aware of how subjective and interpretable quantitative methods are for participants, researchers, and readers, and how large a role quantitative researchers play in generating, analysing and presenting their data. It is not a passive role. It is my view that quantitative methods are no more objective than qualitative methods, qualitative researchers are merely more open and honest about their involvement in the research process. Although I still respect qualitative approaches, I expect I will be more selective in drawing on those approaches than I would have been if I had conducted a qualitative PhD. I am more critical of qualitative approaches that talk about the data ‘emerging’ or ‘giving voice’ to participants. Data does not ‘emerge’ in qualitative or quantitative research. Researchers and participants play an active part in drawing out and analysing data. Researchers do not have the power to ‘give voice’ to participants. Through this PhD, I have come to apply interpretivist and social constructivist concepts to *all* research approaches. This felt in opposition to the post-positivist approaches IDD family research so often appears to be situated in. The longer I spend in the IDD family field however, the more I realise that it is not dominated by post-positivists, it is dominated by pragmatists that are trying to conduct the most robust applied research that they can, whilst underfunded, to provide as much support as possible for people with IDD and their families. I am myself far more of a pragmatist now than I was at the beginning of my PhD studies.

For example, when I began this PhD, I found it very problematic that children and adults with a brother or sister with IDD were referred to as ‘siblings’ even though they were the subject and focus of the work. There is a problem conceptually with the very term ‘sibling’ to refer to people that have a brother or sister with IDD. Even when the sibling is the focus of our study or intervention, they are usually referred to only in relation to their brother or sister with IDD, rather than being referred to as their own person. I have noticed when revisiting earlier empirical chapters in this thesis, for example, that some of my empirical chapters refer to the non-disabled sibling as ‘typically’ developing. The social model of disability highlights the socially constructed notion of ID (cf. Rapley, 2004) and DD. I now

feel that referring to siblings as ‘typically’ developing is not far enough away from referring to them as developmentally ‘normal’. To ease communication, especially when writing about data with comparison groups, it is almost impossible to resist the simplified language of calling this group of participants ‘siblings’, particularly when our work is positioned in the wider IDD field whereby the person with IDD, as they ought to be, is the primary focus. Pragmatically, there is not an attractive alternative to referring to siblings in relation to their brother or sister with IDD, as the term eases communication. We cannot escape, however, that by using the term ‘sibling’ I reinforce the narrative, as all sibling researchers do to some extent, which is to value and care about siblings because of their relationship and caring potential for their brother or sister with IDD. The practical fact remains that this group would not be a focus of our research or interventions if they did not have a brother or sister with IDD, and so now I feel, that to take issue with them being referred to by this relationship, when it is literally the inclusion criteria for focusing on this group of people, is perhaps unnecessarily contrary.

Something that I have come back to throughout this thesis is a desire to disrupt a negative narrative about the effect that having a brother or sister with an IDD can have on a person. I wrote in chapter one about how I have noticed siblings I know and have worked with holding back in order not to advance a negative narrative, and I questioned what this means for our research if participants are withholding information that may help us to further understand their experiences. My empirical chapters have certainly fallen into a negative narrative at times, particularly when situating each empirical chapter and hypothesising why a focus on siblings may be important and necessary. I can see how family systems perspectives can be used both to advance a negative narrative if used over-simplistically, as well as to disrupt it, particularly when focusing on the reciprocal effects siblings can have on one another. We need to avoid pathological assumptions about siblings’ outcomes, because it is disablist at a fundamental level. However, the data I have presented in this thesis do indicate that there are a small group of siblings that may well be at risk of worse psychological outcomes (chapters two and four). Therefore, we cannot ignore that some siblings experience worse psychological outcomes and need support.

What was most interesting about chapters two, four, and five, in my view, was that a straightforward negative narrative about the outcomes of siblings was not substantiated. Once we controlled for other factors the group differences were not statistically significant (chapter two), socio-economic factors as a covariate and as a moderator explained most of the variance for siblings' differing outcomes (chapter four), both children's 'positive' and 'negative' behavioural and emotional outcomes were associated with their sibling relationship in 'positive' and 'negative' ways indicating reciprocal effects at the sibling dyad level (chapter five). If some siblings are experiencing worse outcomes, as the data in this thesis indicates, this appears to be because of indirect factors, and systems based thinking can help us to model future analyses to ensure the complexities and intricacies of siblings' experiences are fully realised by our data in the questions that we ask.

So which siblings are 'at risk'? Unfortunately for me, it seems like it might be siblings like me that are most at risk of worse psychological outcomes. If we think about the findings in chapter two, I have a brother that, as a child, would have scored highly on the SDQ total behavioural problems scale. My mother during my childhood would have scored highly on the K6. We were experiencing high levels of socio-economic deprivation. If we apply family systems perspectives to my family's experience in relation to the findings of that study, we can see how the experiences at the sibling dyad level, and the wider family system, as well as structural factors beyond the family system, were affecting my experiences as a sibling. The importance of socio-economic status in predicting psychological outcomes was a significant finding in this thesis. Rather than feel vindicated that my childhood experiences of poverty were found to be relevant in the data about siblings' outcomes, I feel concerned and somewhat powerless. The overarching solution to supporting siblings is more political and structural in my view. Poverty needs to be eradicated, the world needs to become more equal, and there needs to be comprehensive and extensive governmental support for disabled people and people with mental health support needs throughout the lifespan. People need to stop judging humans' worth based on their ability to contribute economically to society.

I realise that it is naïve and grand to suggest the eradication of poverty and the achievement of equality as something remotely achievable, but in reality, every world problem could be improved by these structural shifts. There are dangers too in

drawing on my personal experiences, for instance, the ambivalence and reciprocity that I found in the final empirical study (chapter five), about sibling relationships, made a lot of sense to me on a personal level. Although this ambivalence is well noted in qualitative sibling studies, it must be noted that I conceptualised the model and tested it, and because it was cross-sectional, it remains purely theoretical, albeit with some data to support its existence. There is always a risk in research that one finds what they choose to find, and we interpret and write in such a way to say what we want to about the world. Perhaps it is this personal experience of the subject matter that makes it so difficult for me to remove myself from the project, even though it is meant to be an 'objective' quantitative study.

Conclusions

My work on this PhD has contributed to new data being generated and analysed about more than one thousand siblings of people with IDD. A strength of this study has been that it has drawn upon nationally representative data and/or large scale survey data to further understand the psychological outcomes of siblings of children and adults with IDD. I have also been able to explore some areas less examined in the existing IDD sibling research, such as sibling relationships and siblings' school experiences. Throughout my studies, I have drawn on family systems perspectives to contextualise, hypothesise and to test the outcomes and experiences of siblings, enabling me – in some ways – to resist a negative narrative about the outcomes of siblings of people with IDD. This work is important because some groups of siblings are in need of specific supports and the findings in this thesis identify areas to concentrate these supports, such as siblings experiencing socio-economic disadvantages. In our social context, this is important, funding for siblings and families is limited, and it is important to focus limited resources on siblings that need it most. Siblings are important as individuals, but also in the lives of their brothers and sisters with IDD. Family systems perspectives enable us to understand that to care about siblings is to care about people with IDD.

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Appendices

Appendix One: Ethics and Study documentation for Chapter Two, Study One

1.1: Expedited Ethical Review Application Form – Final Version



EXPEDITED REVIEW

Application for the Expedited Approval of Research Projects Involving Human (Non-NHS) participants, their data or tissue.

Humanities and Social Sciences Research Ethics Committee (HSSREC)

Expedited review of research can occur where the potential for risk of harm to participants and others affected by the proposed research is minimal, for this reason the review of minor research amendments, research involving only secondary or historical data and research where participant involvement is limited to a consultancy or advisory input may be undertaken via an expedited review process as detailed below:

Please complete sections A and B of this form together with **one** of the annexes and return to the secretary of the HSSREC along with a copy of the study protocol.

Annex A) Approval of minor amendments to projects that have already received Ethical Approval.

Annex B) Approval of research involving the review of secondary or historical data where there is no participant involvement.

Annex C) Approval of research where participant involvement is limited to consultancy/advisory input only.

No research project with ethical considerations may begin before the relevant Sub-Committee of the UREC has issued its written approval. Written confirmation of the Sub-Committee's decision will be emailed to the principal investigator as soon as possible after the expedited review. The Sub-Committee may request a full ethical review for this research project if deemed necessary.

Before completing this form, applicants must refer to the University's Statement and Guidelines on Ethical Practice (<http://www2.warwick.ac.uk/services/rss/>) in conjunction with any other guidance or ethical principles relevant to their specific research.

Section A: GENERAL INFORMATION	
1. Project Title:	Do siblings of children with Learning Disability/Autism in the UK have poorer psychological and educational outcomes?
2. Applicant:	Richard Hastings (PhD supervisor)
3. Department:	CEDAR



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4. Email:	[REDACTED]
5. Telephone	+44 (0)2476 522197
6. Other investigator(s) Institution(s)	The PhD project is part funded by the charity Sibs. Sibs will not be collaborating directly on this study, but they will assist with dissemination of the results.
7. Proposed Start Date	Once ethical approval has been granted.
8. Duration	Until 31 Dec 2018.
9. Funding body	This project is funded through a PhD with the Economic and Social Research Council (ESRC) and additional contributions from the charity Sibs.
10. Are there any potential conflicts of interest?	No If yes, please specify
11. Is this a student project?	Yes If yes, name of student: Nikita Hayden Email [REDACTED]

Section B: DECLARATION

- The information in this form together with any accompanying information is complete and correct to the best of my knowledge and belief and I take full responsibility for it.
- I undertake to abide by the ethical principals underlying the Declaration of Helsinki (<http://www.wma.net/en/30publications/10policies/b3/>) and to abide by the University's Research Code of Conduct (<http://www2.warwick.ac.uk/services/rss/>) alongside any other relevant professional bodies' codes of conduct and/or ethical guidelines.
- If the research is approved, I undertake to adhere to the study protocol without agreed deviation.
- I undertake to inform the HSSREC of any changes in the protocol that would have ethical implications for my research.
- I am aware of my responsibility to be up to date and to comply with requirements of the law and the appropriate guidelines relating to security and confidentiality of participants' personal data.
- Before conducting your research it is strongly recommended that you complete the on-line ethics course:



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https://www2.warwick.ac.uk/services/ldc/researchers/opportunities/development_support/research_integrity/

Support is available from your Departmental contact in Research & Impact Services.

Signature of Principal Investigator

Name (Please Print):Richard Hastings..... Date: 3 May 2017

Signature of Student (if applicable)

Name (Please Print):Nikita Hayden..... Date: 5 May 2017

N.B. For student projects, signatures from both the supervisor and the student are required.

Signature of Chair of Department:

Name (Please Print):Geoff Lindsay..... Date: 4 May 2017

Annex B:

SECONDARY AND HISTORICAL DATA REVIEW (No participant involvement)

1. Please give a brief summary of the project (in lay terms), including the scientific benefit

The overarching research project aims to explore the educational and psychological outcomes for children with siblings with Learning Disability/Autism. The main scientific benefit is the new knowledge generated from the study could inform how to more effectively support siblings. This specific project intends to use the Millennium Cohort Study (MCS) data, a large, UK population based study, to perform a secondary data analysis in order to attempt to answer the question: Do siblings of children with Learning Disability/Autism in the UK have poorer psychological and educational outcomes? By using a UK population-based study we can also provide a clear estimate of any impact of living with a brother or sister with learning disability.

2. Please provide a description of the data source to be used, specifying the era the data relates to and if possible how the data was originally collected.

The Millennium Cohort Study (MCS) is one of the UK's high quality longitudinal cohort studies run by the Centre for Longitudinal Studies currently based at IOE: UCL. Following 19,000 children born



between 2000 and 2001, the study provides insights into both the cohort members and their families. Data are still being collected at specific intervals. This data were created specifically for secondary data analysis and researchers are encouraged to use the data in innovative ways. Participants were fully informed and consented to this.

Due to research already conducted by colleagues in CEDAR (Totsika *et. al.*, 2015), cohort members with learning disability are identifiable. During the second and third waves of MCS (where cohort members are aged three and five respectively), questions were asked related to their older sibling(s) and it is these data, which intends to be explored in this secondary analysis. MCS researchers collected data by visiting households. Sometimes children or parents would self-complete questions using an iPad or computer and at other times the researcher would ask questions, collect data or observe first hand.

3. Please provide details of any ethical review that has taken place regarding the initial collection of the data.

Each wave of data had its own individual ethical approval from the Medical Research Ethics Committee (MREC).

MCS2 – approval granted by the London MultiCentre Research Ethics Committee in September 2004, Reference: MREC/03/2/022

MCS3 – approval granted by the London MultiCentre Research Ethics Committee in December 2005, Reference: 05/MRE02/46 MCS2

MCS3 will be the focus for this specific study.

4. Will approval from another organisation be required in order to gain access to this data, if so please provide the details.

N/A

5. Who will have access to the data?

The data are accessible by any UK researcher agreeing to use data as described by the UK Data Service – as already stated the MCS data have been collected for the purpose of secondary analysis. Data downloaded by the research team will not be shared beyond the team (i.e. other researchers will have to sign up to and agree to the terms set by the UK Data Service to access the data).

6. Please describe any ethical issues/sensitive topics that will be covered during the course of this research.

As a secondary data analysis, information is already anonymized, with no identifying features disclosed to secondary researchers. The main benefit of a secondary data analysis is it will not cause further intrusion in the lives of participants (Dale *et. Al.*, 1988). Participant consent was

1.2: Approval Letter from Ethics Committee



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Humanities and Social Sciences Research Ethics Committee
Kirby Corner Road
Coventry
CV4 8UW

Friday, 26 May 2017

Professor Richard Hastings
CEDAR
University of Warwick
Coventry
CV4 7AL

Dear Professor Hastings,

Ethical Application Reference: 98/16-17

Title: Do siblings of children with Learning Disability/Autism in the UK have poorer psychological and educational outcomes?

Thank you for submitting your expedited application to the Humanities and Social Sciences Research Ethics Sub-Committee for consideration.

We are pleased to advise you that, under the authority delegated to us by the University of Warwick Research Governance and Ethics Committee, full approval for your project is hereby granted.

Before conducting your research it is strongly recommended that you complete the on-line ethics course: https://www2.warwick.ac.uk/services/ldc/researchers/opportunities/development_support/research_integrity/
Support is available from your Departmental contact in Research & Impact Services

Any material changes to any aspect of the project will require further consideration by the Committee and the PI is required to notify the Committee as early as possible should they wish to make any such changes.

May I take this opportunity to wish you the very best of luck with this study.

Yours sincerely



Dr Friederike Schlaghecken
Chair, Humanities and Social Sciences Research Ethics Sub-Committee

Appendix Two: Ethics and Study documentation for Chapter Three, Study Two

2.1: Application for Ethical Approval – Final Version

**HUMANITIES & SOCIAL SCIENCES
 RESEARCH ETHICS SUB-COMMITTEE (HSSREC)**

**Application for Approval of Research Project Involving Human (Non-NHS)
 Participants**

SECTION A. GENERAL INFORMATION	
<p>1. Applicant's Details</p> <p>a. Title:</p> <p>b. Full Name:</p> <p>c. Email:</p> <p>d. Telephone:</p> <p>e. Department:</p> <p>2. Project Title:</p> <p>3. Other Investigator(s):</p> <p>4. Project Start Date:</p> <p>5. Project End Date:</p> <p>6. Funding Body (if any):</p> <p>a. Are there any potential conflicts of interest?</p>	<p>Professor Richard Hastings R.Hastings@warwick.ac.uk 02476 522197 Centre for Educational Development, Appraisal and Research (CEDAR)</p> <p>Evaluation of Sibs Talk school-based intervention for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.</p> <p>Nikita Hayden (PhD student, CEDAR). The research will be conducted in collaboration the charity Sibs.</p> <p>The project will commence once full ethical approval has been given. We anticipate this will be from February 2017.</p> <p>31 December 2018</p> <p>This evaluation is part of a funded collaborative PhD studentship between the University of Warwick's Doctoral Training Centre for the Economic and Social Research Council, and the charity Sibs.</p> <p>No</p>

<p>b. If yes, please specify</p> <p>7. Is this a Student Project?</p> <p>a. Student name</p> <p>b. Student email address</p>	<p>Non-applicable</p> <p>Yes</p> <p>Nikita Hayden</p> <p>N.Hayden@warwick.ac.uk</p>
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Section B: PROJECT DETAILS

1. Please give a brief summary of the project (in lay terms) including the scientific benefit:

The project aims to evaluate the effectiveness of an intervention designed by the charity Sibs. Sibs are the only charity in the UK whose work solely concentrates on supporting the siblings people who are disabled, have special educational needs or a serious long-term condition. Sibs' main aim is to "enhance the lives of siblings by providing them with information and support, and by influencing service provision throughout the UK" (Sibs, 2016). Sibs will be recruiting schools to participate in a pilot project of an in-school support intervention for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition. Laid out in the promotional material (please see appendices 8 and 9) for the interventions, the activities aim to improve siblings' wellbeing and their engagement with learning through:

Normalising the experiences that siblings have growing up in their families;
 Acknowledging the feelings that siblings have about their lives;
 Teaching siblings coping strategies for dealing with difficult situations;
 Enabling schools to make positive changes that enhance siblings' school life.

The training provided by Sibs for schools to lead the intervention also aims to increase the participating schools' knowledge and understanding of this distinct group of pupils. The pupils taking part will be from primary schools. The school-based intervention, hereinafter referred to as Sibs Talk, will consist of:

1. Sibs helping schools to identify siblings in their school
2. Sibs training two school staff members at the school to lead the one-to-one Sibs Talk activities with selected pupils.
3. Schools informing parents of Sibs Talk and getting consent for participants to take part in the evaluation element of the intervention.
4. School staff members explaining Sibs Talk and gaining consent for the evaluation from pupils
5. School staff members leading the activities in Sibs Talk.
6. School staff members will also collect the evaluative data outlined in this ethics form. The researchers will not have direct contact with the pupils or their families.
7. The school will then pass on the evaluative elements of the research, ensuring the data are anonymised before doing so.

A large-scale study by Goudie et al. (2013) suggests that siblings of disabled children may be vulnerable to emotional and behavioural difficulties, and perform less well in terms of educational attainment. Therefore understanding this group of pupils further has a scientific benefit as it will help Sibs, schools and researchers understand this group more, and essentially, help to support them in the most appropriate and effective ways.

Sibs Talk and the evaluation of it are being carried out by Sibs to help them evaluate their school-based work. This ethics request is being put forward to help support Sibs in the analysis and reporting of the evaluation and to ensure that the data are collected appropriately in doing so. By signing up to the free

Sibs Talk resources, the schools agree to the evaluation of Sibs Talk (see appendices 8 and 9) in their partnership agreement with Sibs. By CEDAR being involved in the evaluation, we hope that the research may contribute to wider dissemination of the knowledge generated by Sibs, leading to more impact for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.

2. Please summarise the methodology to be used:

The overarching method being used for this research will be survey questionnaires. Three main questionnaires will be used:

1. A short form for school staff members to complete. This will include basic demographic data such as gender, age, pupil premium status etc. Please see appendix 6.
2. The Strengths and Difficulties Questionnaire (SDQ), including the impact supplement (Goodman, 1999) will be used in the evaluation (please see appendix 3). The pupils' class teachers will complete this questionnaire. It will be completed at the beginning and the end of Sibs Talk. A benefit of this research design approach is that the SDQ has been used for other CEDAR research such as the 1,000 Families Study, as well as for population based studies such as the Millennium Cohort Data (UCL, 2001-2016). Therefore, there are opportunities to compare this group to other sibling cohorts as well as a more general population sample – providing some context for the level of difficulties and strengths that the siblings may have.
3. The "How I Feel About My School" Questionnaire (HIFAMS) (Ford, University of Exeter, 2013 - please see appendix 5) will be employed for the evaluation. The HIFAMS has been designed for 4-11 year old pupils to find out about their subjective attitudes towards school life (University of Exeter Medical School, 2016). The school staff members leading Sibs Talk will sit one-to-one with pupils at the beginning and the end of Sibs Talk and ask them to complete this questionnaire. Guidance on explaining this to the children and gaining consent will be provided for school staff members (please see appendix 4). Some of the older primary school pupils may be able to complete the form with relatively little extra help from the school staff member, whereas those in the lower years groups may need the school staff member to read the questions to the pupil and record their chosen answer. Guidance on this will be included for school staff (please see appendix 4). This questionnaire is a relatively recently published questionnaire that is accessible publically and, over time, may provide bodies of comparable data. The main strength of this questionnaire is that the pupils themselves will provide the information and analysing these data in relation to their teachers' perceptions will provide, we hope, a richer analysis.

In addition, the final activity in Sibs Talk, Activity 10, includes some space for the pupils to make some short comments on the process and their experiences. This information will also be anonymised and shared with Sibs and the Warwick team for the purposes of this study with the permission of the participating pupil (see appendices 7 and 10).

By taking part in the pilot Sibs Talk intervention, schools agree to participate in its evaluation in their partnership agreement with Sibs. As school staff members will be providing pupil data and assisting with the collection of the data, a school staff member guidance document will be provided to assist and ensure school compliance with the ethics requirements outlined in this application.

3. Please describe briefly any ethical issues and / or sensitive topics that will be covered during the course of the project:

Inadvertent distress caused by Sibs Talk (although unlikely for the evaluation itself)

Participants will be asked to reflect on and discuss both positive and negative aspects of being a sibling of somebody who is disabled, has special educational needs or a serious long-term condition. There is a small chance that during Sibs Talk, participants may become upset or highlight an issue that is a safeguarding concern, such as their sibling with, for example, autism, physically harming the pupil at

home. There is no foreseeable risk of harm from participating in the evaluation or Sibs Talk, although risk of inadvertently causing distress within the intervention itself is foreseeable. The evaluation questionnaires themselves are not directly related to children's experiences as a sibling, but are in relation to their schooling and therefore this element of the project is unlikely to cause the child distress when completing it. Evidently any chance of inadvertently causing distress to the participating pupil can be exacerbated by the staff members themselves if they lack sensitivity or are unable to build trust with the participating pupils.

Anonymity, confidentiality and consent

We will be asking school staff to share non-identifiable information about the pupils. However, this still presents issues of confidentiality and anonymity that arise in any project with human participants.

Pupils are primary school aged therefore achieving consent to participate in the evaluation will need to be explained carefully to the children themselves as well as the parents and carers. Sibs Talk and the evaluation will be carried out not by the research team, but by school staff. The age of the participants also presents issues of confidentiality, child protection, safeguarding as well as informed consent challenges.

Power imbalances

Pupils' answers, and verbal assent, could be affected by the power imbalance inherent in any exchange between an adult and child.

Class teachers are also being asked to provide an assessment of the pupils' Strengths and Difficulties (SDQ). Their participation is voluntary, however they may be pressured by the school's Senior Leadership Team to complete this information.

4. How do you intend to handle these areas?

There is a small but foreseeable chance the participants during the Sibs Talk sessions, although unlikely in the evaluation itself, may become distressed or upset in the course of discussing their siblings and the impact of their sibling on their home life and school life. Sibs will be providing training and support to staff members on how to support pupils and how to appropriately manage distress. The training will provide advice on how to sensitively discuss sibling issues and build rapport and trust with the pupils. Sibs will also be able to advise throughout the course of the interventions if issues arise. Sibs have requested that at least one of the two staff members involved in the Sibs Talk interventions in each school are from their Senior Leadership Team, this will insure at least one staff member is very experienced and they can advise and guide the other staff member as well – if needed. For the evaluation part of Sibs Talk, staff members will be given advice both on how to explain the evaluation to the child participating and also how to ethically respond if the child is distressed when completing the information in a school information document. During Sibs Talk itself, if a child becomes distressed or if a safeguarding issue occurs - the safeguarding procedures developed by individual schools will be followed. School staff members would have received training at the school clarifying this. This will be reinforced by the Sibs trainers.

In terms of anonymity, data will be collected by school staff members and passed on, anonymised, to Sibs, and then passed on to University of Warwick researchers based in CEDAR to analyse – again in an anonymised format. Staff members will be advised on how to collect this information (please see appendices 4 and 7). Parents will have already been informed about Sibs Talk by the school and given the opportunity to remove their child from participating in Sibs Talk. Parents will then be sent information about the evaluation and the research and asked to "opt-out" if they do not want their child's data and evaluation responses to be shared with Sibs and the researchers (please see return slip at the end of appendix 1). Although the children are of primary school age, they will be provided with information about

the study and asked to assent verbally to participating.

Pupils' answers could be affected by the power imbalance inherent in any exchange between an adult and a child. Staff members will be given training by Sibs and will be advised to build rapport with the pupils to help create a relaxed and comfortable space for the Sibs Talk sessions and evaluation of it. Those leading Sibs Talk will be asked to reiterate to the pupils that their answers will be kept confidential and they should answer as honestly as possible, explaining anonymity and confidentiality in a way the child can understand. This will hopefully encourage pupils to answer the HIFAMS questionnaire as openly as possible. It is acknowledged, however, that the adults' influences and the power imbalance may still impact the research.

School staff will be asked to complete the SDQ. By completing the form they consent to the information being used in the way described in their participant information sheet (see appendix 2). Their completion of this information is voluntary: they retain the right to refuse to participate. However, they will be being asked to complete this information by a member of their SLT – therefore a power imbalance may influence their decision to complete the information. Their voluntary participation will be highlighted and emphasised in both the school information, which the SLT reads, as well as their participant information sheet. To minimise the possibility of any adverse consequences happening due to a teacher refusing to participate, they will be provided with an envelope to put the questionnaires in and to seal. Therefore, when the teacher passes this on to the SLT lead – they will not know if the forms have been completed. This is to reduce significantly the chance of adverse consequences from refusing to take part, ensuring that participation is voluntary.

5. What possible risks are there for the researcher?

The researcher will not be collecting the data first-hand. There are no identified risks for the researchers named in this project in relation to this research.

6. Will you, or any of the research team, who will come into contact with participants be required to obtain criminal record clearance?

No

7. If answered 'yes' to 6, please confirm that such clearance will be obtained:

Non-applicable

SECTION C: PARTICIPANTS

1. How will participants be recruited?

Participants will be recruited through their schools. Sibs is working collaboratively with schools and recruiting new participating schools to take part in the pilot project. The senior leadership team at the school, by taking part in the Sibs Talk, agree to support the evaluation of Sibs Talk – as illustrated by the schools' Sibs Talk information sheet from Sibs (see Appendices 8 and 9) and a partnership agreement. Two staff members will be trained in leading the Sibs Talk intervention and the evaluation of Sibs Talk, one of which will be a member of the senior leadership team who will act as a "gatekeeper" to pupils, ensuring the consent of parents has not been withdrawn and assent from the pupils has been obtained before information is shared with Sibs or the researchers.

2. How many participants will be recruited?

50-60 pupils (estimated)

3. How will informed consent be obtained from the participants (please provide a consent form and participant information sheets to be used)? If no consent will be obtained please explain why:

Parents and carers of potential participants will be sent an information sheet detailing the study and will be offered an opportunity to opt-out their child from the research evaluation. The parents will be informed that the following anonymised data will be shared with the researchers and Sibs:

1. The school will share anonymised demographic data about their child (please see appendix 6)
2. Their child will be asked to complete a questionnaire at the beginning and the end of Sibs Talk and share their answers from the Sibs Talk activity booklet (please see appendices 4, 5, 7 and 10)
3. Their child's class teacher will complete a questionnaire about their child at the beginning and the end of Sibs Talk (please see appendices 2 and 3)

The parent can withdraw their child from the evaluation by contacting their child's school. Using an opting out strategy for this research is appropriate because this is a very low risk evaluation focused on support for pupils that the schools will be putting in place whether or not the evaluation takes place. In addition, it is crucial to obtain evaluation data to inform future roll outs of Sibs Talk and opting in consent procedures with parents reduce significantly the response rate for school based research – leading to biased evaluations of the interventions. We also want to ensure that the consent process does not exclude unnecessarily those pupils who may have more complex home situations, who may be even less likely to respond to opting in consent procedures. Finally, because the data will only be passed over to the researchers and Sibs in an anonymised format, we feel that this provides another safeguard for pupils and parents, which further supports the use of an opt-out for parental consent to pupil participation in this evaluation.

The participating child will be given information verbally by the school staff member leading Sibs Talk and asked if they understand and agree to take part in the evaluation. A script will be provided for the school staff member (please see appendices 4 and 7) to help explain the evaluation to the child. The information will clarify that the child should not be coerced or forced to take part in the evaluation and their involvement in the evaluation is voluntary – it should not impact on their ability to take part in Sibs Talk if they, and their parent, wishes that they do so. The child will be asked if they understand the research and will be given the opportunity not to participate if they do not want to.

School class teachers will be asked to complete a SDQ for each child taking part on Sibs Talk. Teachers will be given an information sheet to read. They consent to the SDQ by completing it and passing it back on to the school lead Sibs contact in a sealed envelope. If the teacher does not consent, then they pass back the incomplete questionnaires to the SLT lead in the sealed envelope provided. The information sheet will clarify the material needed to gain informed consent from class teachers (see appendix 2). It

will be emphasised both to the SLT and themselves that their consent and completion of the information is voluntary and they may refuse to participate. We hope this will remove the issue of a power imbalance resulting from the SLT asking a class teacher to complete documentation that needs to be completed voluntarily.

4. Will deception be used during the course of the research?

No

5. If yes, why is it deemed necessary?

Non-applicable

6. Will the participant group include any children or vulnerable adults?

Yes

7. If yes, please explain the necessity of these individuals:

The Sibs Talk intervention has been designed to support primary school aged pupils who have a sibling who is disabled, has special educational needs or a serious long-term condition. The evaluation is being used to improve this intervention as well as understand the experiences of this specific group in greater detail. Asking pupils to self-report on their experiences and attitudes is key to understanding this cohort further. This element of the evaluation adds richness and resists a simplistic narrative, which excludes the voices of those at the heart of the research. It may have been possible to merely ask parents and teachers to provide ratings about the pupils. However, this would give a more limited perspective. Involving the children themselves in the research allows those at the centre of the project to have their perspectives heard, giving a more varied view of the interventions and their changing experiences. This could not be achieved without their involvement.

8. If yes, please also explain how and from whom fully informed consent will be obtained:

As the participants are children, parents and carers of potential participants will be sent an information sheet detailing the study and will be offered an opportunity to opt their child out from the research evaluation. The parents will be informed of the nature of the data being collected as well as the purpose. They will be informed that fully anonymised data will be shared with the researchers and Sibs (please see appendix 1). The parent can withdraw their child from the evaluation by contacting their child's school.

The participating child will be given information verbally by the school staff member leading Sibs Talk and asked if they understand and agree to take part in the evaluation. As the pupils are primary school aged, a verbal explanation would be preferable over a written explanation at this stage. A script will be provided for the school staff member (please see Appendices 4 and 7) to help explain the information to the child. The child will be asked if they understand the research and will be given the opportunity not to participate if they do not want to. Thus, pupils will provide assent to take part in the research evaluation.

9. Will participants be given payment and/or incentives for participating in the research?

No

10. If yes, please specify level of compensation and source of the funds or incentives. Please also explain the necessity of such compensation:

Non applicable

11. What possible benefits and/or risks to participants are there to this research?

Pupils gain an opportunity through Sibs Talk to reflect on their experiences and learn more about themselves and their circumstances. They will develop ways to understand and communicate their emotions and experiences. By sharing their experiences with their parents/carers and the school staff member leading Sibs Talk, these adults may develop ways to better support those children participating in the study and children like them.

The evaluation will inform the work of Sibs to help them develop Sibs Talk and improve the support they offer to siblings of people who are disabled, have special educational needs or a serious long-term condition. Research findings can be shared in an accessible format with the participating schools specifically for them to disseminate to parents and pupils. This will further support these pupils at school and at home, informing future interventions and best practice for schools.

There is a risk that participants may become distressed through participating in Sibs Talk – and staff members leading Sibs Talk will be advised and trained on how to manage this if the situation arises. Schools will have their own procedures and policies for this. There is no other identified risk of harm for pupils. Furthermore, for the evaluation component, there is a near zero risk of inadvertently causing distress for the pupil.

12. What arrangements have been made for reporting the results of the research to and/or debriefing the participants:

The final Sibs Talk session has been designed to allow feedback and to re-explain the evaluation to the participating pupils. The research evaluation findings will be shared in an accessible, anonymised and aggregated format to the school for them to share with the parents and pupils involved in the research. No findings will be shared or published in a way that could identify any of the participants. Sibs and the researchers will not have any pupil identifiable data. Sibs will know which schools participated – but they will not pass on this specific information to the researchers writing an evaluation report. Sibs will also publish a version of the evaluation report.

13. What qualified personnel will be available to deal with possible adverse consequences/reactions to those participating in this research?

It is highly unlikely that there will be any adverse consequences from taking part in the research. A strength of the research design is that the research will be carried out at the school where school personnel are trained and equipped to manage any adverse consequences relating to Sibs Talk. A staff member will be leading Sibs Talk and the pupils' class teacher (who will know the pupil well) will be on site and will be informed of any issues that arise. In addition, the senior leadership team can support if a safeguarding issue arises and parents can be informed of any concerns by the school. Furthermore Sibs provides an online platform for participants to access information and support – this service is highlighted on the back of the Sibs Talk booklet and in training with those leading Sibs Talk. Sibs staff members are also available for schools to contact for information and advice on responding appropriately to siblings if issues arise.

SECTION D: DATA

1. How will you ensure confidentiality? *(Please give details of how and at what stage in the project you will anonymise the data):*

Data will be passed on to the research team already anonymised by the participating schools' senior leadership teams. The schools will already be well-versed in data protection and confidentiality requirements and will also be reminded of this in the training delivered by Sibs and information sent to them about the study. Therefore the researchers will receive a confidential version of the data. No identifying information, including the names of schools, will be passed to the University team. All research data will also be stored in password-protected computer folders which only the researchers will have access to. We will be advising schools in a schools' information document on how to handle and pass over the documents confidentially. Once anonymised versions of the data have been passed on to Sibs and the University of Warwick, we will require the schools to destroy any remaining personal data they hold on the evaluation of the Sibs Talk intervention.

2. Who will have access to the data?

Researchers from CEDAR and staff from Sibs will receive anonymised versions of the data. The participating schools will destroy data relating to the evaluation once anonymised versions have been passed on to Sibs and CEDAR staff.

3. Where will consent forms, information sheets and project data be stored?

Consent forms will not be collected for this project as parents will be invited to opt-out of the research (and their opt-outs would be retained/destroyed confidentially by the participating school) and children will be asked to provide verbal assent. All research project data will also be stored in password-protected computer folders which only the researchers will have access to.

4. For how long will the above data be kept and how and when will data then be destroyed?

The anonymised survey data will be securely stored for a minimum of 10 years, in line with University of Warwick policy on secure university servers in a password protected computer that only the researchers will have access to. The school will be advised to destroy evaluation-related data once anonymised versions have been passed over to Sibs and CEDAR.

5. Is it anticipated that there will be any future use of the data and have the participants been informed of this use?

None anticipated beyond the current proposed evaluation of Sibs Talk and its dissemination as described in the participant information sheets.

6. Will any interviews be audio or video taped? If yes, please attach a copy of the consent/authorisation form.

Not applicable

SECTION E: PUBLICATIONS

1. How will publications of research findings recognise the contributions of all researchers engaged in the study?

All researchers will be recognised as co-authors on any resulting publication or report. The order of names will reflect input and will be agreed by the team.

SECTION F: FURTHER INFORMATION

1. Please give any additional information you believe to be relevant to this project:

To help safeguard against staff members not following our ethical guidelines described in this application, we will require the lead contact to confirm they have read and adhered to our ethical requirements (please see Appendix 11).

Citations:

Ford, U. o. E. (2013). "'How I Feel About My School' (HIFAMS) questionnaire." Retrieved 15-12-2016, 2016.

Goodman, R. (1999). "The extended version of the Strengths and Difficulties Questionnaire as a guide to child psychiatric caseness and consequent burden." *Journal of Child Psychology Psychiatry* **40**: 791-799.

Goudie, A., et al. (2013). "Assessing Functional Impairment in Siblings Living With Children With Disability." *Pediatrics* **132**(2): e476-e483.

Sibs (2016). "About Sibs: Aims, history, strategy." *Sibs website*. Retrieved 15-12-2016, 2016.

Appendices:

- A1 Parent PIS
- A2 Class teacher PIS
- A3 SDQ English(UK)_t4-17full (1)
- A4 HIFAMS questionnaire script and staff information
- A5 HIFAMS Questionnaire
- A6 Demographic information form
- A7 Assent to share Activity 10 guidance
- A8 About Sibs Talk handout
- A9 Siblings in Schools Project – information for school staff
- A10 Sibs Talk Activity Booklet
- A11 Confirmation of ethical procedures for evaluation data collection

2.2: Signed Declaration for Ethical Approval – Initially Submitted Version

SECTION G: DECLARATION

The information in this form together with any accompanying information is complete and correct to the best of my knowledge and belief and I take full responsibility for it.

I undertake to abide by the ethical principles underlying the Declaration of Helsinki (<http://www.wma.net/e/policy/b3.htm>) and to abide by the University's Research Code of Conduct (<http://www2.warwick.ac.uk/services/rss/>) alongside any other relevant professional bodies' codes of conduct and/or ethical guidelines.

If the research is approved, I undertake to adhere to the study protocol without agreed deviation.

I undertake to inform the HSSREC of any changes in the protocol that would have ethical implications for my research.

I am aware of my responsibility to be up to date and to comply with requirements of the law and the appropriate guidelines relating to security and confidentiality of participants' personal data.

Signature of Principal Investigator ...

Date: 17/1/17

Signature of Student (If Applicable) ..

Date: 17/1/17

Signature of Head of Department

Date: 17/1/17

Please send an electronic copy of the application to HSSREC@warwick.ac.uk.
Note. Your electronic submission should contain wet signatures of all relevant parties.

2.3: Participant Information Sheet – Parent Version

PARTICIPANT INFORMATION SHEET
Parent on behalf of pupil invited to take part
version 4, 08/02/2017

Study Title: Evaluation of Sibs Talk school-based intervention for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.

Investigator(s): Richard Hastings (University of Warwick), Nikita Hayden (University of Warwick), Claire Fraser-Lim (Sibs), Monica McCaffrey (Sibs)

Introduction

As you are already aware, your child has been invited to take part in Sibs Talk activities during school time to help support them. Sibs Talk consists of ten intervention activities between a school staff member and your child. This is because your child has been identified as someone who has a brother or sister who is disabled, has special educational needs or a serious long-term condition. To ensure that Sibs Talk is as useful as it can be for your child and other siblings, the charity Sibs and the University of Warwick will be evaluating it. To evaluate the Sibs Talk intervention, four pieces of information will be collected about your child: (i) Your child's class teacher will complete a questionnaire at the beginning and end of the Sibs Talk intervention; (ii) A school staff member will ask your child to answer some questions before and after the Sibs Talk intervention about school, if they would like to; (iii) A school staff member will ask your child if they would like to share a page summarising their experiences from their Sibs Talk booklet; (iv) Anonymised demographic information about your child e.g. gender, age, ethnicity etc.

Your child's information will be included in the evaluation unless you send the enclosed form to their class teacher to withdraw your child from the evaluation. This information will be anonymous. This means that the information Sibs and the University of Warwick will be given will not make you or your child identifiable in any way. We will not be able to find out your names, who you are, where you live etc. Before you decide, you need to understand why the research is being done and what it would involve for you and your child. Please take the time to read the following information carefully. Talk to others about the study if you wish.

(Part 1 tells you the purpose of the study and what will happen to you and your child if you take part. Part 2 gives you more detailed information about the conduct of the study)

Please contact Claire Fraser-Lim, Development Officer for Siblings in Schools, if there is anything that is not clear or if you would like more information. Claire can be contacted at: Address: Sibs, Meadowfield, Goose Green, Oxenhope, West Yorkshire, BD22 9JD.

Email: schools@sibs.org.uk

Telephone: 01535 645453

Take time to decide whether or not you wish for your child to take part.

PART 1

What is the study about?

This study will be used to evaluate how useful the Sibs Talk intervention has been for pupils.



Does my child have to take part?

It is entirely up to you to decide by reading this information sheet and asking follow up questions if you choose to do so. If you choose to allow your child to be involved in the evaluation of the Sibs Talk intervention for pupils, you do not need to do anything. Your child will be asked at the beginning of the intervention if they want to take part and, if they agree, then your child's anonymised information will be included in the evaluation study. If you do not want your child to take part in the evaluation please inform your child's class teacher using the form enclosed. You will be free to withdraw your child from the evaluation study at any time before the final Sibs Talk intervention session. After this, the information will be passed on anonymously to Sibs and the University of Warwick. To withdraw your child from the evaluation you do not have to give a reason and this will not affect your child or their circumstances in any way.

What will happen if my child takes part?

You should have already received some information from the school about Sibs Talk. In addition, your child is being asked to take part in an evaluation of the Sibs Talk intervention. If you decide not to allow your child to take part in the evaluation element, your child can still take part in Sibs Talk if you would still like him/her to. Your child will be asked to complete a short questionnaire at the start of the first activity and at the end of the final activity. Your child will be told about the research and asked if they are happy to share this information. They will also be asked to share some short comments on their views about the Sibs Talk intervention, but they do not have to if they do not want to. The class teacher will also be asked to complete a questionnaire at the beginning and at the end of the activities about your child. This will help us learn more about what your child is like at school and if this changes as a result of the activities. The school staff member who is doing the activities with your child will complete some basic information about your child such as their gender, their age and whether they have a disability etc. The school will make sure that your child cannot be identified by this. Your child's name will be removed from any documents passed on to Sibs: no identifiable data will be shared with Sibs or the University of Warwick. The information will be used to evaluate the Sibs Talk intervention. The research may be disseminated at conferences and in PhD research and publications. All data obtained for the evaluation will be reported on an aggregated basis – this means that we will report on all the information together – so your child and their answers will not be individually reported on. Names of pupils, teachers and schools will not be used so your child will not be identifiable.

What are the possible disadvantages, side effects, risks, and/or discomforts of your child taking part in this study?

Your child will give up a small amount of time to take part in the evaluation of the Sibs Talk intervention that they receive at school. There are no other identified disadvantages or risks associated with taking part in this evaluation.

What are the possible benefits of taking part in this study?

Taking part in this study will allow us to find out more information to evaluate a support intervention that may benefit pupils who have a brother or sister who is disabled, has special educational needs or a serious long-term condition at home.

Expenses and payments



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We will not be offering payments for taking part in this study. Taking part in this study will incur no expenses for you or your child.

What will happen when the study ends?

At the end of the study, the questionnaires and forms will be shared with Sibs and the University of Warwick in an anonymised format. This will then be securely stored for a minimum of 10 years, in line with University of Warwick policy on secure university servers in a password protected computer file which only the researchers will have access to. A summary report of the research will be provided to your school and shared with you. You or your child will not be identifiable in this report, nor any publications or presentations that are produced from it.

Will my taking part be kept confidential?

Yes. We will follow strict ethical and legal practice and all information about you will be handled in confidence. Further details are included in Part 2.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm that you might suffer will be addressed. Detailed information is given in Part 2.

This concludes Part 1.

If the information in Part 1 has interested you and you are considering taking part, please read the additional information in Part 2 before making any decision.

PART 2

Who is organising and funding the study?

This study is being conducted in collaboration with Sibs, a charity that supports children and adults who are siblings of people who are disabled, have special educational needs or a serious long-term condition and the Centre for Educational Development, Appraisal and Research based at the University of Warwick. This evaluation is part of a funded collaborative PhD studentship between the University of Warwick's Economic and Social Research Council Doctoral Training Centre, and the charity Sibs.

What will happen if I do not want my child to take part in this study?

Taking part in this study is entirely voluntary. Deciding not to take part will not affect you or your child in any way.

If you decide later that you do not want your child to take part in the evaluation project, you may withdraw your child from the evaluation research at any time without it affecting you or your child in any way.

Who should I contact if I wish to make a complaint?

Any complaint about the way you or your child has been dealt with during the study or any



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possible harm you or your child might have suffered will be addressed. Please address your complaint to the person below, who is a senior University of Warwick official entirely independent of this study:

Head of Research Governance

Research & Impact Services
University House
University of Warwick
Coventry
CV4 8UW
Email: researchgovernance@warwick.ac.uk
Tel: 024 76 522746

Will my child's taking part be kept confidential?

The anonymised survey data will be securely stored for a minimum of 10 years, in line with University of Warwick policy on secure university servers in a password protected computer file. Data will be destroyed in accordance with the University's procedures at that time. Your child's information will be kept confidential at all times including when sharing the findings of the study. Your child's name and school's name will not be used in any resulting reports, papers or presentations and will remain confidential.

What will happen to the results of the study?

The results of the study will be used to evaluate Sibs Talk. Findings of the research will be shared with schools that take part. The schools will also be encouraged to pass on this information to parents and pupils. The findings may also be used anonymously in academic conferences and publications to help researchers further understand this group of pupils and their psychological and educational experiences.

Who has reviewed the study?

This study has been reviewed and given favourable opinion by the University of Warwick's Humanities and Social Science Research Ethics Committee (HSSREC): **Insert your HSSREC reference number here (given to you when your study is approved) and include the date on your approval letter from HSSREC.**

What if I want more information about the study?

If you have any questions about any aspect of the study - or your child's involvement - not answered by this information sheet, please contact:

Primary researcher - Nikita Hayden, University of Warwick, [REDACTED]
Project lead for Sibs - Claire Fraser-Lim, Sibs, schools@sibs.org.uk; Research supervisor - Richard Hastings, University of Warwick, [REDACTED]

Thank you for taking the time to read this Information Sheet.

✂.....

Sibs Talk evaluation - for pupils who have a brother or sister who is disabled or has special educational needs or a serious long-term condition



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If you would **not** like your child's information to be shared in the way described in this information sheet, please return this completed slip to your child's class teacher.

Pupil's name: Pupil's class:

I **would not like** my child to take part in the evaluation of the Sibs Talk intervention. Please do not send my child's anonymised information to Sibs or the University of Warwick.

*If you **would like** your child to take part in the evaluation then you do not need to return this slip or do anything. Your child's anonymised information will be included in the evaluation.*

Signed: Date:

Study Two: Participant Information Sheet – Class Teacher Version



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PARTICIPANT INFORMATION SHEET

Class teacher

version 4, 08/02/2017

Study Title: Evaluation of Sibs Talk school-based intervention for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.

Investigator(s): Richard Hastings (University of Warwick), Nikita Hayden (University of Warwick), Claire Fraser-Lim (Sibs), Monica McCaffrey (Sibs)

Introduction

As you have already been informed by the school's senior leadership team, some pupils at the school have been identified as children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition and have been invited to take part in a pilot study of an intervention (Sibs Talk) to help support them.

The Sibs Talk intervention has been developed by the charity Sibs who, alongside the University of Warwick, are working to find out more about ways we can support and help this group of children. To help with this, the intervention is being evaluated as part of a research project. We would like to ask you, as a participating pupil's class teacher, to complete the attached Strengths and Difficulties Questionnaire (SDQ) for each child participating in both the intervention *and* evaluation of Sibs Talk. Therefore, please do not complete this questionnaire for pupils whose parent/carer has informed you via return slip that they would like to withdraw their child from the evaluation element of Sibs Talk.

We ask that you complete this questionnaire just before each child starts the first Sibs Talk intervention session, and again after the final session. In total, there are ten session activities that the participating pupils will complete as part of the Sibs Talk intervention. We hope this evaluation will achieve two things. In the short-term, this research will help Sibs evaluate and improve their in-school intervention programme and approach. Secondly, it will inform research currently being conducted in collaboration between the University of Warwick and Sibs to learn more about the educational and psychological experiences of siblings of disabled children and learn how best to support them.

Your completion of the SDQ about each child taking part in the Sibs intervention, as well as the evaluation, is completely voluntary. Before you decide if you want to take part, you need to understand why the research is being done and what it would involve for yourself. Please take the time to read the following information carefully. Talk to others about the study if you wish.

(Part 1 tells you the purpose of the study and what will happen to you if you take part. Part 2 gives you more detailed information about the conduct of the study)

Please contact Claire Fraser-Lim, Development Officer for Siblings in Schools, if there is anything that is not clear or if you would like more information. Claire can be contacted at: Address: Sibs, Meadowfield, Goose Green, Oxenhope, West Yorkshire, BD22 9JD.

Email: schools@sibs.org.uk

Telephone: 01535 645453



Take time to decide whether or not you wish to take part in this study.

PART 1

What is the study about?

This study will be used to evaluate how useful the Sibs Talk intervention has been for pupils.

Do I have to take part?

It is entirely up to you to decide by reading this information sheet and asking follow up questions if you choose to do so. If you choose to participate, you will be asked to complete the Strengths and Difficulties Questionnaire about each pupil that is eligible to participate i.e. taking part in both the Sibs Talk intervention *as well* as the evaluation. If you do not want to participate, please do not complete the questionnaires. The school lead and SLT at the school have been informed that class teacher consent in this research is completely voluntary and that there should be no consequences for you if you decide not to complete the SDQ. You are free to withdraw your consent at any time, without giving a reason and this will not affect you or your circumstances in any way. If you do not consent, please place the uncompleted forms into the envelope(s), seal them, and return them to the SLT Sibs contact. If you do not take part, no other staff member will complete the SDQ and the questionnaire will be returned uncompleted to the researchers.

What will happen if I take part?

You should have already received some information from the school about the Sibs Talk intervention. If you take part, you will be asked to complete a SDQ for each child in your class who takes part in the Sibs Talk intervention at school. The forms should take no more than five minutes per pupil before the intervention starts and again at the end. The SDQ focuses on pupils' strengths and difficulties in things like their social skills, and what sort of impact you think these strengths and difficulties have for the pupil at school.

Once you have completed the forms or have read this information and decided not to, you will then need to pass this information back to the Sibs Talk intervention lead contact in the SLT. Please place the questionnaires into the white envelope provided before passing them back to the SLT contact – this way your participation or declination to participate in the study will be kept confidential. This information will then be passed back to Sibs charity and the researchers – with no pupil identifying information being passed on. The information will be used to evaluate the Sibs intervention. The research may be disseminated at conferences, and in PhD research and publications. All data obtained for the evaluation will be reported on an aggregated basis - names of pupils, teachers and schools will not be used. Your SLT will know you were asked to complete the SDQ for certain pupils, however, they will be asked to treat this information confidentially. They will not know whether or not you completed the forms as you will return them in a sealed envelope.

What are the possible disadvantages, side effects, risks, and/or discomforts of you taking part in this study?

You will be giving up a small amount of time to complete the questionnaires for each pupil. There are no identified disadvantages or risks associated with taking part in this research evaluation.

What are the possible benefits of taking part in this study?

Participating in this study will allow us to generate information to evaluate a support intervention



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that may benefit pupils who have who have a brother or sister who is disabled, have special educational needs or a serious long-term condition.

Expenses and payments

We will not be offering payments for taking part in this study. Taking part in this study will incur no expenses for you either.

What will happen when the study ends?

At the end of the study, the questionnaires will be shared with Sibs and the University of Warwick in an anonymised format. This will then be securely stored for a minimum of 10 years, in line with University of Warwick policy on secure university servers in a password protected computer file which only the researchers will have access to. A summary report of the research will be provided to your school and shared with you. You will not be identifiable in this report, nor any publications or presentations that are produced from it.

Will my taking part be kept confidential?

Yes. We will follow strict ethical and legal practice and all information about you will be handled in confidence. Further details are included in Part 2.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm that you might suffer will be addressed. Detailed information is given in Part 2.

This concludes Part 1.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

PART 2

Who is organising and funding the study?

This study is being conducted in collaboration with Sibs, a charity created specifically to support children and adults who have a brother or sister who is disabled, has special educational needs or a serious long-term condition and the Centre for Educational Development, Appraisal and Research based at the University of Warwick. This evaluation is part of a funded collaborative PhD studentship between the University of Warwick's Economic and Social Research Council Doctoral Training Centre, and the charity Sibs.

What will happen if I do not want to take part in this study?

Participation in this study is entirely voluntary. Refusal to participate will not affect you in any way.

If you agree at this stage to participate, you may nevertheless withdraw from the evaluation research at any time without it affecting you in any way.

Who should I contact if I wish to make a complaint?



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Any complaint about the way you have been dealt with during the study or any possible harm you might have suffered will be addressed. Please address your complaint to the person below, who is a senior University of Warwick official entirely independent of this study:

Head of Research Governance

Research & Impact Services
University House
University of Warwick
Coventry
CV4 8UW
Email: researchgovernance@warwick.ac.uk
Tel: 024 76 522746

Will my taking part be kept confidential?

Your data will be securely stored for a minimum of 10 years, in line with University of Warwick policy on secure university servers in a password protected computer file which only the researchers will have access to. Data will be destroyed in accordance with the University's procedures at that time. Your information, and that of the pupil participant, will be kept confidential at all times including when sharing the findings of the study. Your name, the pupil's name and school's name will not be used in any resulting reports, papers or presentations and will remain confidential.

What will happen to the results of the study?

The results of the study will be used to evaluate the Sibs Talk intervention which has been designed to help children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition. The findings of the research will be shared with participating schools. The schools will also be encouraged to pass on this information to parents and pupils. The findings may also be used anonymously in academic conferences and publications to help researchers further understand this group of learners and their psychological and educational experiences.

Who has reviewed the study?

This study has been reviewed and given favourable opinion by the University of Warwick's Humanities and Social Science Research Ethics Committee (HSSREC): **Insert your HSSREC reference number here (given to you when your study is approved) and include the date on your approval letter from HSSREC.**

What if I want more information about the study?

If you have any questions about any aspect of the study not answered by this participant information sheet, please contact:

Primary researcher - Nikita Hayden, University of Warwick, [REDACTED]

Project lead for Sibs - Claire Fraser-Lim, Sibs, schools@sibs.org.uk

Research supervisor - Richard Hastings, University of Warwick, [REDACTED]

Thank you for taking the time to read this Information Sheet.

2.4: Strengths and Difficulties Questionnaire – School version

Strengths and Difficulties Questionnaire

T 4-17

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months or this school year.

Pupil code

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, cannot stay still for long	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often complains of headaches, stomach-aches or sickness	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often has temper tantrums or hot tempers	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, tends to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally obedient, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries, often seems worried	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Helpful if someone is hurt, upset or feeling ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Constantly fidgeting or squirming	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Has at least one good friend	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often fights with other children or bullies them	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often unhappy, down-hearted or tearful	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Easily distracted, concentration wanders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Nervous or clingy in new situations, easily loses confidence	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Kind to younger children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often lies or cheats	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Picked on or bullied by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often volunteers to help others (parents, teachers, other children)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Thinks things out before acting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Steals from home, school or elsewhere	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Gets on better with adults than with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many fears, easily scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Sees tasks through to the end, good attention span	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Do you have any other comments or concerns?

Please turn over - there are a few more questions on the other side

Overall, do you think that this child has difficulties in one or more of the following areas: emotions, concentration, behaviour or being able to get on with other people?

No	Yes- minor difficulties	Yes- definite difficulties	Yes- severe difficulties
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

If you have answered "Yes", please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties upset or distress the child?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties interfere with the child's everyday life in the following areas?

	Not at all	Only a little	Quite a lot	A great deal
PEER RELATIONSHIPS	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
CLASSROOM LEARNING	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties put a burden on you or the class as a whole?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Thank you very much for your help

2.5: HIFAMS Questionnaire Script and Staff Information

Example script for school staff members to explain the HIFAMS questionnaire to the participating pupil

This script and information has been adapted from the HIFAMS website: Ford, University of Exeter (2013).

Completing the HIFAMS – guidance for those leading the Sibs Talk intervention

To help us evaluate the Sibs Talk intervention, participating pupils should complete the HIFAMS questionnaire at the beginning and end of the intervention. The questions are designed to evaluate the intervention booklets. Pupils should not be coerced or forced to complete the evaluation, it should be a voluntary process. We ask you to help the participating pupil understand this evaluation and to complete this evaluation at the beginning and at the end of the first and last intervention sessions.

We have provided a script to help you explain the questionnaire to the pupil (adapted from the HIFAMS website). You may find that you need to explain it slightly differently depending on the age and reading level of the child you are working with.

Key Stage 2

For the Sibs Talk intervention, the HIFAMS should be completed on a one-to-one basis with the child. Once you have run through the introduction, give the child a questionnaire to complete and make yourself available to assist if the child is struggling with the questionnaire. Depending on the level of the pupil you are working with, you may need to run through some examples with the pupil. You may also need to read each question to the pupil and help them circle the appropriate face, or even do this for them. Please use your own discretion – dependant on the comprehension of the child. Once the questionnaire has been completed, please check that the child has completed the questionnaire correctly, circling only one face per question.

Outline script

Please note: We do not intend this script to be read verbatim. There is room to change the exact wording if necessary. The structure should remain the same though, and the finalised examples of happy/neutral/sad responses should not be changed between pupils.

"Today, we'd like you to help us with a special project. We're going to ask you some questions about school and how school makes you feel. The answers you give us will be used to help other children like you who have a brother or sister who is disabled, has special educational needs or a serious long-term condition. Your answers will be kept very safe and private – as your name won't be used – just your answers.

"There are no right or wrong answers; we just want to know how you feel. It is very important you answer the questions on your own without my ideas or opinions. We really want to hear what you yourself have to say.

"To answer the questions you need to circle the face you think shows how you feel. If you feel happy, circle the happy face; if you're unhappy, circle the sad face; if you're neither happy nor unhappy, circle the OK face. Let's do some practices now:

"How does going to a party make you feel?

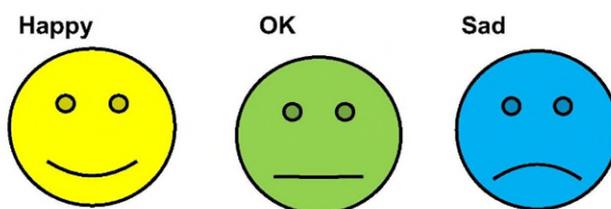
"How does falling over in the playground make you feel?

[Use other examples as needed]

"Do you understand? If your answers make us worried about you, we might have to talk to someone who can help us to help you.¹

"If you don't want to take part anymore then just let me know and we can stop."

Example faces from the questionnaire



¹ If applicable, please refer to your own policies related to safeguarding and explaining confidentiality to your pupils. This will ensure you explain confidentiality to your pupils in the most appropriate way for your setting.

2.6: HIFAMS Questionnaire

Pupil code

How I Feel About My School

Instructions:

Please put a circle around the face that shows how you feel.

	Happy	OK	Sad
On my way to school I feel ...			
When I am in the classroom I feel ...			
When I am doing my work I feel ...			
When I am in the playground I feel ...			
When I think about the other children at school I feel ...			
When I think about my teacher I feel ...			
When I think about school I feel ...			

Thank you very much for doing this 

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2.7: Demographic Information Form

Sibs Talk Key Stage 2 evaluation

Please complete the following information about each child taking part in the evaluation.

A. About the pupil

1. Pupil code

2. Age in years: 3. Gender:

4. Is this pupil categorised as:

4a. Attracting Pupil Premium funding Yes No

4b. Speaking English as a Second Language Yes No

4c. Disabled or identified as having Special Educational Needs Yes No

5. Please select which ethnicity the pupil is categorised as:

White - British	<input type="checkbox"/>	Pakistani	<input type="checkbox"/>
White - Irish	<input type="checkbox"/>	Bangladeshi	<input type="checkbox"/>
Traveller of Irish Heritage	<input type="checkbox"/>	Any other Asian Background	<input type="checkbox"/>
Any other White Background	<input type="checkbox"/>	Black Caribbean	<input type="checkbox"/>
Gypsy/Roma	<input type="checkbox"/>	Black – African	<input type="checkbox"/>
White and Black Caribbean	<input type="checkbox"/>	Any other Black Background	<input type="checkbox"/>
White and Black African	<input type="checkbox"/>	Chinese	<input type="checkbox"/>
White and Asian	<input type="checkbox"/>	Any other Ethnic Group	<input type="checkbox"/>
Any other Mixed Background	<input type="checkbox"/>	Refused	<input type="checkbox"/>
Indian	<input type="checkbox"/>	Information not yet obtained	<input type="checkbox"/>

6. Please describe what disability definition the pupil's brother/s or sister/s has/have (i.e., name the condition)

.....

B. About the Sibs Talk intervention

7. What was the date of the first intervention session?

8. What was the date of the final intervention session?

9. Out of 10 possible intervention sessions, how many were completed in total?

10. What is the role/ position of the school staff member who led the intervention with this child?

.....

Thank you for completing this information.

2.8: Assent to Share Activity 10 Guidance

Sibs Talk evaluation

Assent from participating pupil to share Activity 10 from their booklet - guidance for school staff member leading the Sibbs Talk intervention

Part A: Information about pupil assent to share Activity 10 from their Sibbs Talk booklet

To help us evaluate the Sibbs Talk intervention, participating pupils should be asked if they would be happy to share their answers from their Sibbs Talk work booklets for Activity 10. We ask that you carefully explain what this means to the pupil using the guidance and script included. Informed assent should be gained from the pupil before it is photocopied and shared with Sibbs and the University of Warwick. This part of the verbal assent should be completed before the pupil completes Activity 10. Once Activity 10 has been completed, you then complete the HIFAMS questionnaire with the pupil again. Please revisit and reflect on previous activities with the pupil when they are completing Activity 10 to help them recall previous activities as well as the more recent ones. This also provides an opportunity for the pupil to reflect on and review the sessions.

Gaining a photocopy of their completed Activity 10 will contribute to our evaluation of the Sibbs Talk intervention booklets. Pupils should not be coerced or forced to share this information, it should be a voluntary process which is why we ask you to gain the pupil's assent *before* they complete the information. We ask you to help the participating pupil understand what the photocopied page will be used for. With their assent, this will be shared with Sibbs and the University of Warwick to help us understand their experiences and help other children who have brothers and sisters who are disabled, have special educational needs or a serious long-term condition.

We have provided a script to help you explain this to the pupil. You may find that you need to explain it slightly differently depending on the age and comprehension of the pupil you are working with: the script does not have to be spoken verbatim, although the structure should remain similar.

Script to explain sharing their work to the pupil:

“The people who made this booklet we have been using want to find out about how useful the booklet was for you. We are about to do the last activity and before we do, I would like to find out if you are happy to share your answers to Activity 10 with the people who made the booklet and the people who are trying to find out more about siblings like you. If you say yes, I will photocopy this page once you have finished filling it out [point to/show the child Activity 10]. This will be used to help other children like you. Your answers in your booklet will still be kept very safe and private – as your name won't be used – just your answers.

“There are no right or wrong answers; we just want to know what you think. We really want to hear what you yourself have to say.

Sibs Talk evaluation**Assent from participating pupil to share Activity 10 from their booklet - guidance for school staff member leading the Sibbs Talk intervention**

"We are now going to go through some of your previous activities to remind you what work you have done. This may remind you of things you want to share in Activity 10. You will then write your answers for this activity. Are you happy to share your answers?"

"If you don't want to take part anymore - just let me know and we can stop."

Please proceed to review the activities and help the pupil complete Activity 10. Sharing their answers to Activity 10 is voluntary - if the pupil does not want to share their answers, you should still complete the activities but please do not provide us with a copy of their answers.

Please ask follow up questions to encourage the pupil to think about the ten activities – not just the most recent. Check the pupil understands the way the information will be used by asking questions. Once Activity 10 has been completed, check again that the pupil is happy to share their answers. Please complete the following form for each pupil.

.....

Sibs Talk evaluation – assent to provide photocopies of activity 10 from the Sibbs Talk activity booklet

Pupil code

1. I confirm that I have explained to the participating pupil how a photocopy of Activity 10 from their Sibbs Talk booklet will be shared if they give their assent (please tick to confirm).
2. The participating pupil has agreed to allow me to photocopy and pass on Activity 10 (please attach a photocopy of Activity 10)
3. The participating pupil has not agreed to share a copy of Activity 10 (please do not provide a photocopy of Activity 10)

Signed:

Date:

2.9: About Sibs Talk Handout

Sibs

talk

Sometimes I get embarrassed when people stare at my sister.



I'm glad my brother is like he is. I love playing games with him.



A booklet for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition.

Sibs

For brothers and sisters
of disabled children and adults

Sibs Talk is for pupils who have a brother or sister who is disabled or has special educational needs or a serious long-term condition.

Many schools are working with pupils who are having problems at school because of their sibling experiences.

Sibs Talk has been designed to help schools support these siblings. It is a one-to-one support intervention for school staff to use with siblings in Key Stage 2. Its purpose is to improve siblings' wellbeing and their engagement with learning.

Each page of the booklet is a short session for the sibling to work through with a member of staff. There are ten sessions that can be spread out over a term.

The activities in the booklet aim to:

- Normalise the experiences that siblings have growing up in their families
- Acknowledge the feelings that siblings have about their lives
- Teach siblings coping strategies for dealing with difficult situations
- Enable schools to make positive changes that enhance siblings' school life.

Sibs Talk links in with the following policies and initiatives in schools:

- Developing wellbeing and resilience
- Identifying young carers
- Celebrating difference and diversity
- Anti-bullying and safeguarding
- SEND support.

Sibs Talk is part of a research project being evaluated by the University of Warwick as it is important that schools have an evidence-based approach to supporting siblings. The **Sibs Talk** package includes staff training, a staff instruction manual, activity booklets for siblings and the evaluation materials. **It is free to a limited number of primary schools.**

To arrange a phone call to discuss **Sibs Talk** email schools@sibs.org.uk

www.sibs.org.uk
 info@sibs.org.uk  01535 645453
 Sibs_uk  SibsCharity

Registered charity no. 1145200 Limited company no. 7834303

2.10: Siblings in Schools Project – Information for School Staff

Sibs

For brothers and sisters
of disabled children and adults

New Siblings in Schools Project

Help us identify and support children and young people in your school that are growing up with a brother or sister who is disabled or has special educational needs or a serious long-term condition.



Who We Are

Sibs is the only UK charity dedicated to supporting siblings of people who are disabled, have special educational needs or a serious long-term condition.

There are over **half a million children** and young people in the UK who are growing up with a brother or sister with these needs. **That's almost two children in every school classroom.**

Young siblings are vulnerable to isolation, bullying, reduced wellbeing and problems with attainment. They are more likely than their peers to experience public prejudice, family breakdown and bereavement.

Over half of all young carers in the UK are siblings of disabled children.

Four out of ten siblings live in poverty.

"School would have been so much easier if my teachers had known about my sister Katie who is non-verbal and autistic.

Growing up we shared the same room so my sleep was interrupted when she had a bad night.

That made it hard to concentrate in lessons the next day. My homework - when I had a chance to do it - was regularly scribbled on and when things were difficult at home it was hard to keep my emotions in check.

There were so many times I was in need of support or attention at school, but nobody knew."

Laura, sibling.



For more information
visit www.sibs.org.uk

Why This Issue Matters To Schools

Many children are either not identified as siblings or have their **needs overlooked** leading to problems with progress at school.

Sibling young carers have significantly **lower educational attainment** at GCSE level and are more likely than the national average to be not in education, employment or training (NEET) between the ages of 16 and 19.

Our aim is that every school in the UK will know how many of its pupils have a brother or sister who is disabled or has special educational needs or a serious long-term condition.

You may already be working with pupils who are having problems at school **because of their sibling experiences.** Barriers to learning for siblings include disturbed sleep and helping with care. Siblings may require specialist help such as counselling or support with homework. They may also need extra information and guidance at key points in their school life.

Schools can also help siblings celebrate the many positives around being a sibling such as developing patience, tolerance and kindness.

"We want siblings of disabled children and those with special educational needs or a life-long serious illness to have every opportunity to progress and achieve at school and beyond.

The first step towards achieving this goal has to be for schools to know how many of their pupils are siblings."

Monica McCaffrey, Chief Executive,
Sibs



What You Can Do



Take part in our exciting new Siblings in Schools Project to help us find out what works for siblings in schools and what helps them progress.

Start by contacting our Development Officer for Siblings in Schools to register your interest email schools@sibs.org.uk

What we can do to support your school.

We will:

- Advise you on how to identify the siblings that attend your school
- Provide our new activity booklets for all of these siblings
- Train two members of staff on how to use these booklets with siblings
- Support siblings at your school through our YoungSibs online service
- Collect data about sibling wellbeing and attainment with the University of Warwick
- Evaluate the use of the booklets and provide you with a report on this
- Involve you in publications about best practice on supporting siblings in schools.

We will provide the above support and expertise FREE to a limited number of pilot schools. If you want to develop a sibling support group in school, there is a charge for sibling group leader training.

What commitment do we expect from your school?

We want you to:

- Identify a minimum of 10 siblings to work through the new activity booklets
- Agree to two staff members leading on the work, one of whom needs to be on SLT
- Allocate two hours of staff time for training on using the booklets
- Provide Sibs with the data to evaluate the effectiveness of the project
- Work within the project timescales set in agreement with Sibs.

www.sibs.org.uk

✉ info@sibs.org.uk
 🐦 [Sibs_uk](https://twitter.com/Sibs_uk)

☎ 01535 645453
 📘 SibsCharity

Registered charity no. 1145200 Limited company no. 7834303

2.11: Confirmation of Ethical Procedures for Evaluation Data Collection

Confirmation of ethical procedures for evaluation data collection

To be completed by the Sibs Talk lead contact

Please read and complete the following declaration at the end of the Sibs Talk interventions when you are ready to pass over the anonymised data for all participating children in your school. We ask you to complete this form once in relation to the data for all of the children for whom you are sending the data. Please send Monica McCaffrey this form along with the evaluation data to Monica McCaffrey, Sibs, Meadowfield, Goose Green, Oxenhope, West Yorkshire, BD22 9JD.

Part A

I confirm that I have read and followed the instructions provided by Sibs and the University of Warwick to ensure the data being passed on has been collected ethically.

This information has been provided in the following documents:

1. In the training session with Sibs
2. In the instruction manual provided by Sibs
3. In the How I Feel About My School (HIFAMS) script and staff information
4. In the guidance relating to assent to share activity 10

Part B

I confirm I, or another school staff, member has:

1. Provided each participating child's parent/carer with the parent information sheet before the evaluation of Sibs Talk commenced
2. Removed any children's data if their parent/carer has withdrawn their consent for them to take part in the evaluation
3. Ensured that staff delivering the Sibs Talk intervention fully understand the ethics of data collection for this evaluation
4. Ensured that staff delivering the Sibs Talk intervention have asked participating children for their assent to share their data from page 10 of the Sibs Talk booklet
5. Ensured that staff delivering the Sibs Talk intervention have asked participating children for their assent to share their data from the HIFAMS questionnaires
6. Ensured that children's class teachers have been provided with the information sheet about the evaluation and that their involvement has been voluntary with regard to the completion of the Strengths and Difficulties Questionnaires
7. Anonymised any pupil data using the codes provided by Sibs before passing them on to Sibs
8. In addition, I confirm that once the anonymised data have been passed on to Sibs, I will destroy any personal data relating to the evaluation of Sibs Talk. I have kept no copies and am sending on the original data forms

Full name

Signed Date

2.12: Approval Letter from Ethics Committee



WARWICK
THE UNIVERSITY OF WARWICK

Humanities and Social Sciences Research Ethics Committee
Kirby Corner Road
Coventry
CV4 8UW

Wednesday, 15 February 2017

Professor Richard Hastings

CEDAR
University of Warwick
Coventry
CV4 7AL

Dear Professor Hastings,

Ethical Application Reference: 41/16-17

Title: Evaluation of Sibs Talk school-based intervention for children who have a brother or sister who is disabled, has special educational needs or a serious long-term condition

Thank you for submitting your updated ethics application to the Humanities and Social Sciences Research Ethics Sub-Committee, following the letter of conditional approval on **01 February 2017**. We are pleased to advise you that, under the authority delegated to us by the University of Warwick Research Governance and Ethics Committee, full approval for your project is hereby granted for the duration of the study.

Before conducting your research it is strongly recommended that you complete the on-line ethics course: https://www2.warwick.ac.uk/services/ldc/researchers/opportunities/development_support/research_integrity/
Support is available from your Departmental contact in Research & Impact Services

Any material changes to any aspect of the project will require further consideration by the Committee and the PI is required to notify the Committee as early as possible should they wish to make any such changes.

May I take this opportunity to wish you the very best of luck with this study.

Yours sincerely



Dr Friederike Schlaghecken
Chair, Humanities and Social Sciences Research Ethics Sub-Committee

Appendix Three: Ethics and Study Documentation for Chapter Four, Study Three

3.1: Application for Ethical Approval – Final Version



**Humanities & Social Sciences Research Ethics Committee (HSSREC):
Application Form for Research Ethical Approval**

Date: 20.06.2019	Version: 1
SECTION 1. APPLICANT DETAILS	
1.1 APPLICANT	
Applicant's Title (optional):	Ms
Applicant's Forename:	Nikita
Applicant's Surname:	Hayden
School or Department:	Centre for Educational Development, Appraisal and Research
Warwick e-mail address:	[REDACTED]
Contact telephone number:	[REDACTED]
Applicant's Status:	
STUDENT:	STAFF:
Undergraduate Student <input type="checkbox"/>	Professor <input type="checkbox"/>
Taught Postgraduate Student <input type="checkbox"/>	Associate Professor <input type="checkbox"/>
Postgraduate Research Student <input checked="" type="checkbox"/>	Assistant Professor <input type="checkbox"/>
Name of course/qualification:	Research Fellow <input type="checkbox"/>
PhD in Psychology and Education	Teaching Fellow <input type="checkbox"/>
	Other <input type="checkbox"/>
	Please specify:
1.2 SUPERVISOR (COMPLETE FOR ALL STUDENT PROJECTS)	
Supervisor's Title:	Professor
Supervisor's Forename:	Richard
Supervisor's Surname:	Hastings
Supervisor's Post:	Professor and Cerebra Chair of Family Research
Supervisor's Faculty/School and Department:	Centre for Educational Development, Appraisal and Research
Supervisor's Warwick e-mail address:	[REDACTED]
Supervisor's contact telephone number:	[REDACTED]
1.3 OTHER INVESTIGATORS/COLLABORATORS (INTERNAL & EXTERNAL)	
Please list all other known collaborators, internal and external to Warwick, including the name of the company/organisation or Investigator's Warwick department/school and their role in the project:	
Nikita Hayden's PhD is a collaborative award between the ESRC, the University of Warwick and the UK charity Sibs. Sibs staff, and trustees, who are also adult siblings themselves, suggested topics of inquiry to include and provide feedback on the survey. Sibs will help to publicize the survey through their mailing list	

and social media presence.

Alison Baker, administrator, Centre for Educational Development, Appraisal and Research, University of Warwick

1.4 REFERRALS

Has the Project been referred to HSSREC from another REC or delegated process? No
If yes, please provide the reason:

Referred by department as not within the remit for delegated approval

Other

Please provide details:

SECTION 2. PROJECT DETAILS

2.1 Project Title:	Adult sibling survey
2.2 Estimated start date:	September 2019
2.3 Estimated completion date of project:	September 2020
2.4 Does the project involve the NHS or social care:	No
2.5 Type of Project: https://warwick.ac.uk/services/ris/research_integrity/researchethicscommittees/biomed/study_design/ Research <input checked="" type="checkbox"/> NHS Service evaluation or Development <input type="checkbox"/> NHS Clinical Audit <input type="checkbox"/> Other- please specify:	
2.6 Research Sponsor: If not research in the NHS, please state N/A	N/A
2.7 Funder: If unfunded, please state N/A	This study is part of a PhD project that is funded by the Economic and Social Research Council and the UK charity Sibs
2.8 IDEATE/Funder reference (if applicable) If your study is funded, please provide a reference	Economic and Social Research Council grant ES/J500203/1
2.9 Links with other HSSREC applications Is the project linked to any other HSSREC application? No If yes, detail: Project title: Chief Investigator: HSSREC Reference (if known): Nature of linkage:	

SECTION 3: BACKGROUND/LAY SUMMARY

Please provide a lay summary of the project:
 The summary should be brief and easily understood by someone who is not an expert in the area. Definitions and explanation of terms should be provided (avoid technical language).
To include:

- a description of the proposed study and population to be studied building on review of previous studies/evidence
- the scientific benefit of the proposed study

Context and literature review

It is often thought that siblings of individuals with intellectual or developmental disabilities (IDD) may have different experiences and outcomes than their peers with a typically developing brother or sister. Life expectancies have improved for people with various conditions due to advances in care and treatment (for example people with Down syndrome; Hodapp & Urbano, 2007). Increasing life expectancies of those with disabilities may also result in siblings being expected to take on caring and support roles for their siblings when their parents are no longer able to. Furthermore, the wellbeing of siblings is important to value and promote even if siblings do not have the desire or capacity to provide care and support for their disabled brothers and sisters. Previously Davys et al. (2011) and Heller & Arnold (2010) have investigated the literature on adult siblings who have a brother or sister with a learning disability or developmental disability, respectively. Their work highlights that the available evidence about these adult siblings is limited. Where sibling research about children has grown in recent decades, the evidence exploring adult siblings has remained relatively small and limited.

There is evidence to suggest some adult siblings are at a greater risk of psychological problems. Rai et al. (2018) used a Swedish population-based cohort study with 223,842 participants to explore depression in young adults aged between 18 and 27 years old (mean age 21.5), comparing depression in young adults with and without Autism Spectrum Disorders (ASD). They also found that the adult siblings of people with ASD were at an increased risk of depression than the adult siblings of people without an ASD. Other large-scale studies have not found that adult siblings were at a greater risk of worse psychological outcomes, finding quite similar outcomes for siblings of people with and without disabilities (specifically mild intellectual disabilities and mental illness – Taylor et al. 2008). The differences in the results are likely to be due to a range of design differences, however it may also be in part due to the age of the participants. Rai et al. had a cohort of young adults, whereas Taylor et al. used a cohort of adults in middle age. Therefore a sample that involves adult siblings from across the life stages may help us to understand how sibling experiences and outcomes change over time, and not just in a particular life stage. We also know for instance, the disability of the sibling with an IDD is important, as adult siblings of people with Down syndrome seem to have better psychological outcomes and better sibling relationships than siblings of people with ASD (Hodapp and Urbano, 2007; Orsmond & Seltzer, 2007). Further work is needed to understand adult siblings' psychological wellbeing, risk factors, and differences relating to poverty, ethnicity and religion.

Relatively few studies have concentrated on adult sibling relationships where one has an ID, however, a stronger more positive relationship between siblings and their brother or sister with a disability have been associated with the sibling providing higher levels of support and assistance for their brother or sister (Tomeny et al., 2017). Therefore, those interested in the future care and wellbeing of disabled adults would perhaps be interested in ways of improving and fostering the relationship between siblings and their brother or sister with a disability. Sibling relationships are also important because they are potentially the longest relationship a person will have, typically lasting across the life-span and arguably helping to form the basis of many social behaviours (Mandleco & Webb, 2015). For individuals with ASD and ID, their relationship with their siblings may be even more important, particularly if they have fewer peer relationships and rely on family members primarily for friendship, care and support (Richardson & Jordan, 2017). Overall though, sibling relationships appear to be fairly positive where one sibling one has an intellectual disability (ID) or a developmental disability (DD; Hodapp et al. 2010). We know however, that some groups of adult siblings have better relationships than others, and fostering positive relationships, and understanding how to do this, is important. There are a few suggested protective factors for sibling relationships including, being from a larger family (Hodapp et al., 2010), having a brother or sister with fewer challenging behaviours (Orsmond et al., 2009), or having a brother or sister with Down syndrome (Orsmond & Seltzer, 2007; Hodapp & Urbano, 2007) as opposed to more profound or severe ASD (Doody et al., 2010), or both siblings being of the same sex (Orsmond et al., 2009). Studies have identified the need for support in siblings of people with disabilities (Arnold et al., 2012). However, there are few studies that help us understand what this support would need to consist of.

Current study

The current study seeks to further understand the experiences, wellbeing and the needs of adult siblings of people with IDD as well as their sibling relationships with one another. An online survey (hosted on the

Qualtrics platform, on the approved list of HSSREC suppliers and the Information and Security team) will be administered (see appendix 5) to adult siblings of people with IDD in the UK. The UK charity Sibs will share the link to siblings in their network. The survey will be shared on social media, and IDD charities will also be encouraged to share the link in order to reach further families (see email appendix 4 and poster in appendix 3). The online survey has been carefully designed to ensure that appropriate measures have been selected as far as possible so there are data to compare our findings to. When selecting measures, we have selected pre-existing, robust measures with high internal consistency and good psychometric properties.

Scientific benefit

Increasing life expectancies of people with IDD as well as the reduction in welfare and increasing poverty rates in the UK indicate that siblings may become increasingly important in the lives of their brothers and sisters with IDD. We seek to understand the wellbeing outcomes of adult siblings of people with IDD and their relationships with their brothers and sisters with IDD. We want to explore adult siblings' caregiving role or their expectations for caring in the future, as well as the needs of siblings, so that we can consider ways to support their wellbeing and support the role they decide to take in their brother or sister with IDD's life. There are some data to suggest that some adult siblings of people with IDD may be at an increased risk of psychological problems. They therefore may be an at-risk group in themselves, due to the direct or indirect stressors of having a brother or sister with IDD. Understanding this risk and considering ways to support siblings is therefore also important.

SECTION 4 RISK AND ETHICAL CONSIDERATIONS CHECKLIST

Complete the checklist ticking 'Yes' or 'No' to **all** questions.

Where you have ticked 'Yes' to a question below, you will need to specifically address the ethical issues raised by that point and detail what safeguards will be put in place to minimise the potential risks/harm in the relevant section of the application form or in the space provided.

		Yes	No
A	Does the study involve participants who are particularly vulnerable or unable to give informed consent or in a dependent position (e.g. children, your own students, over-researched groups, people with learning difficulties, people with mental health problems, young offenders, people in care facilities, prisoners)? <i>(If yes, please provide details in section 7 – Informed Consent)</i>	<input type="checkbox"/>	<input checked="" type="checkbox"/>
B	Will participants be taking part in the study without their consent or knowledge at the time, or will deception of any sort be involved (e.g. covert observation of people in non-public places)? <i>(If yes, please provide details in section 7 – Informed Consent)</i>	<input type="checkbox"/>	<input checked="" type="checkbox"/>
C	Is there a risk that the highly sensitive nature of the subject might lead to disclosures from the participant concerning their involvement in illegal activities or other activities that represent a threat to themselves or others (e.g. sexual activity, drug use, or professional misconduct)? If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
D	Could the study induce psychological distress or anxiety , or produce humiliation , or cause harm , or lead to negative consequences beyond the risks encountered in normal life? <ul style="list-style-type: none"> • <i>Applicable to studies involving sensitive topics, vulnerable participants as well as studies involving driving experiments, simulators, computational or physiological experiments. For the latter, please detail potential risks associated with any equipment and how these will be monitored and addressed in the space below.</i> 	<input type="checkbox"/>	<input checked="" type="checkbox"/>

	<ul style="list-style-type: none"> Please also consider the risk to individuals if any personally identifiable data collected as part of the study is accidentally disclosed. Please see guidance note for more information. <p>If yes, please provide details:</p>		
E	Does the study involve substantial physical exertion ? If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
F	Does the study involve the administration of any substance? If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
G	Does the study involve physically intrusive procedures , use of bodily materials or human tissue , or DNA/RNA analysis ? If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
H	Is any reward , apart from travelling and other expenses, to be given to participants? If yes, please provide details and justification for this, to ensure this is appropriate, and not seen as a bribe or to coerce participants into taking part:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
I	Could the proposal give rise to researchers having any conflicts of interest ? https://warwick.ac.uk/services/finance/resources/regulations/fp1 <ul style="list-style-type: none"> Consider relationships/previous personal interactions with participating organisations, participants etc. <p>If yes, please provide details including how this will be managed:</p>	<input type="checkbox"/>	<input checked="" type="checkbox"/>
J	Will any part of the project be undertaken overseas? If yes, please state which Country/Countries, the locations at which the project will be undertaken, e.g. public place, school, company, hospital, University, researcher's office, including the services of an overseas cloud hosting provider for storage or a market research company etc. and the local permissions in place for this (where required): Please see University Guidance for data processing overseas: https://warwick.ac.uk/services/idc/dataprotection/internationaldatatransfers/	<input type="checkbox"/>	<input checked="" type="checkbox"/>
K	Will the researchers go to any areas where their safety may be compromised ? If yes, please provide details, including what measures will be put in place to minimise risks and ensure the researcher's safety. A risk assessment should be submitted with the application:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
L	Will pregnant women be participants in the study? <ul style="list-style-type: none"> Please note, while you may not purposefully be recruiting pregnant women to the study, consider if any special measures would need to be put into place or if it is appropriate for these individuals to take part, e.g. safety risks <p>If yes, please provide details:</p>	<input type="checkbox"/>	<input checked="" type="checkbox"/>
M	Will the study involve children under 5 years old? If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
N	Is the research commissioned by the military ?* If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
O	Is the research commissioned under an EU security call ?* If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
P	Does the research involve the acquisition of security clearances ?* If yes, please provide details:	<input type="checkbox"/>	<input checked="" type="checkbox"/>
Q	Does the research concern terrorist or extreme groups ?*	<input type="checkbox"/>	<input checked="" type="checkbox"/>

	If yes, please provide details:		
R	Does the study involve any additional ethical considerations or risks to participants or the researcher that are not listed above?	<input type="checkbox"/>	<input checked="" type="checkbox"/>
	If yes, please provide details:		

* Please refer to the University webpages on [Prevent Duty](#)

SECTION 5: STUDY DESIGN, METHODOLOGY & ANALYSIS

5.1 Clearly state the research aim(s) of the project:

To include:

- a clear explanation and justification for the research question(s)/aim(s)

The aim of this survey is to further understand the wellbeing and outcomes of adult siblings of people with IDD, their relationships with their brothers and sisters with IDD, their caregiving role or expectations for caring in the future, as well as the needs of siblings.

We ask the following research questions:

1. What are the psychological, social and health outcomes for adult siblings of people with IDD?
2. What care and support role do adult siblings of people with IDD have towards their brothers and sisters with IDD?
3. How do adult siblings with IDD perceive their relationship with their brother or sister with IDD?
4. What information and support needs do adult siblings of people with IDD have?
5. What factors predict or are associated with these outcomes?

5.2 What are the objective(s) for the project:

- Objectives are intermediate steps that will help you to meet your research aim(s)

The immediate steps to be taken to meet our research aims include the following:

1. To develop a survey measuring psychological, social and health outcomes, sibling relationships, carer burden and support needs, as well as familial, disability and socio-demographic information, in order to capture the outcomes required to explore the above research questions.
2. To recruit to this survey study using our existing collaboration with the UK charity Sibs, as well as with other UK IDD support charities and social media.
3. To make efforts to recruit a large, diverse sample of adult siblings to maximise analysis opportunities. By diverse, we mean recruiting adult siblings from across the life-span and recruiting adult siblings from lower-socioeconomic backgrounds etc.

5.3 Study design and data collection methods:

To include:

- a clear description of the study design and data collection methods
- a suitable design should reflect the aim(s) of the study
- This may include ethnography/observations, interviews, focus groups, questionnaires, document analysis etc.
 - **Ethnography/Observations**- what/who will be observed, by whom, for how long? What equipment (if any) will be used for recording etc.?
 - **Interviews**- who is conducting the interviews, how, where and when- by telephone/in person/skype; will they be recorded- how? How long will they last? How will the interview guide be developed? etc.
 - **Focus groups**- who is leading, how will they be organised, when and where will they take place, how will they be recorded? How long will they last? etc..
 - **Questionnaires**- who has designed the questionnaire, who will distribute it, how long will it take to complete etc.
 - **Document analysis**- what documents will be requested, where from, by whom, what permissions are in place for this etc.
 - **Experimental** – what tests/lab work will be undertaken on participants, by whom, is specialist training required before undertaking?

- **Secondary analysis of previously collected data-** *analysis of data that has been previously collected by a third party for research or other purposes, that is not publicly available e.g. healthcare, student, financial records. Please state whether the data set is identifiable or anonymised.*

An online survey (hosted on the Qualtrics platform, on the approved list of HSSREC suppliers and the Information and Security team) will be administered (see appendix 5) to adult siblings of people with IDD in the UK. The UK charity Sibs will share the link to adult siblings in their network. The survey will also be shared on social media, and IDD charities will also be encouraged to share the link in order to reach further families (see email appendix 4 and poster in appendix 3). Adult siblings will be directed to a survey hosted on the University-approved survey platform Qualtrics, and asked to read the information sheet (appendix 1) and to tick and agree to the consent form items (appendix 2). They will then begin the online survey (appendix 5). The survey will be fully anonymous – we will not collect any identifying information.

The online survey has been carefully designed to ensure that appropriate measures have been selected as far as possible so there are data to compare our findings to. For instance, we have selected questions and options from national data such as the Census data questions (Census forms, 2011) or questions from the 'Survey of Informal Carers in Households 2009/10' (2010). Questions about the socioeconomic position of families mirror questions asked in the population study, the Millennium Cohort Study (MCS, 2019), allowing further comparisons to be made between adult siblings of people with IDD, the general population, as well as other carer groups and other disability groups. When selecting measures, we have selected pre-existing, robust measures with high internal consistency and good psychometric properties. For example, we will measure the wellbeing of adult siblings using measures from Kessler (2003) and WEMWBS (2019) and we will measure the sibling relationship using a reduced measure from Riggio (2000). These not only indicate the reliability and robustness of the selected measures, but they also provide comparisons to be made about siblings of people with IDD to "normative" populations as well as to adult siblings of people with other disabilities – as some of these measures have been used with studies of other co-related populations. We have also consulted with colleagues in the United States and Canada who are currently collecting data for adult siblings of people with IDD in their respective nations to consider ways of comparing the findings of our study to the findings of their studies. Furthermore, our collaboration with the UK charity Sibs, the only charity in the UK with the sole purpose of supporting siblings of people with disabilities, provides further feedback on the relevance and appropriateness for this group of participants. The survey will take approximately 20 to 25 minutes to complete. Measures and items have been removed to avoid overlapping lines of inquiry. We have also significantly reduced the number of items in measures – for example, the Lifespan Sibling Relationship Scale (Riggio, 2000) has been reduced by half – removing the retrospective childhood items – so that the questions asked are focused on answering our research questions.

5.4 Data Analysis

To include:

- *Specifically what data sets will be collected (name, date of birth, email address, ethnicity, health status, financial records, IP addresses, etc.)*
- *whether this data will be collected directly from participants (e.g. via questionnaires/interviews) or indirectly, from a third party (previously collected data set) and how i.e. web form, online application, paper form*
- *Detail the analysis methods that will be undertaken e.g. content analysis, framework analysis, interpretative phenomenological analysis etc. and any statistical analyses.*
- *Describe how and by whom any data will be transcribed, coded, de-identified, stored, transferred, accessed, archived*
- *Any software used in the analysis should be specified and detailed how it will be used in the project*

Data will be collected directly from participants through an online survey. The full survey (appendix 5) details all the data to be collected from participants. There are some sensitive data about disability, ethnicity, financial information and age. However, this information is anonymous and all questions will be optional. Therefore, participants can skip questions they do not wish to answer. We will be collecting postcodes so that we can extract data from the census about neighbourhood deprivation. Once the deciles of neighbourhood deprivation have been assigned to each case in the data set, the postcodes will be removed and stored in a separate folder and destroyed after 10 years. All files will be on a password protected, encrypted University of Warwick device and only accessible to the University of Warwick team.

Data will be analysed using multivariate analysis techniques (using SPSS). Data from the final, open text question will be analysed using content analysis by hand (rather than using qualitative analysis software).

The data will be downloaded weekly from Qualtrics by the research team. These copies of the data will be

password protected, encrypted and saved on the M drive in a folder only the research team based at the University of Warwick will have access to. No identifying information is being collected therefore the data is anonymous.

SECTION 6: RECRUITMENT

6.1 State the total number of planned participants and the sampling strategy; provide justification for this:

To include:

- *The rationale behind the proposed size of the sample*
- *Will the sample size provide enough data to answer the research question?*
- *If sampling will be continued until saturation is reached, then this should be stated and linked to the research question*
- *Sampling strategy- is this random, snowball, purposive, convenience etc.*
- *What is the rationale for this- it should reflect the methodological framework for the study*

We aim to recruit more than 200 adult siblings to take part in this study, although we would continue to recruit adult siblings over this number (up to 2,000 adult siblings). The sampling strategy would involve convenience sampling through the UK charity Sibs as well as other disability organisations and through social media. We will encourage participants to share the survey with their other siblings or anyone else they know who is a sibling of someone with an IDD, therefore using snowball sampling. A challenge in recruiting adult siblings in this way is that this may result in a homogenous sample, and this may make including ethnicity, gender and socio-economic position for example, not feasible. For instance, Sibs has noticed that white women disproportionality attend their events. However, they have a very large mailing list – and this may consist of adult siblings that are more representative of the UK population, but who are unable or do not want to access face-to-face Sibs' services. The socio-demographic characteristics of early participants will be reviewed and steps will be taken towards a more targeted approach to recruitment if necessary, for example, by contacting local disability service organisations in neighbourhoods with high proportions of black, Asian and minority ethnic people or in neighbourhoods with high socio-economic deprivation.

6.2 Where applicable, state the breakdown of participants by type and number of each type of participant, e.g. children (include age), parents, teachers, health care professionals etc.:

Type of Participant:	Number:
Adult sibling of people with IDD	200-2,000

6.3 Please provide clear inclusion criteria:

- The participant must be aged 18 years or over.
- The participant must have a brother or a sister with an intellectual or developmental disability.
- The participant must reside in the UK.
- The participant must be willing to take part in the research.

6.4 Please provide clear exclusion criteria:

- The participant is under 18 years of age.
- The participant does not have a brother or a sister with an intellectual or developmental disability.
- The participant does not reside in the UK.
- The participant is not willing to take part in the research.

6.5 Please detail **how** participants will be recruited to the study:

To include:

- *How participants will be identified/screened and approached; by whom?*
- *Where participants will be recruited from and when?*
- *Detail the source of any personal information that may be used to identify participants. If this information will be accessed by someone outside the team who would have access to this information as part of their day to day role, the reason for this should be explained, and permissions detailed e.g. healthcare, student records etc.*
- *Will any vulnerable groups be recruited?*
- *What materials will be used to recruit participants- please provide copies of posters, leaflets, invitation emails, etc.*

- *Where will the above materials be advertised: list and provide details of locations, websites, social media etc.*
- *Will any recruitment tools be used e.g. SONA- please specify and provide details.*

A Qualtrics link containing the survey will be shared (see email in appendix 4) with organisations that work with families of people with IDD. We will encourage these organisations to share a poster (see appendix 3) to their mailing list and to their social media accounts. The UK charity Sibs is our collaborative partner on this study and they will also promote the study with the siblings they work with. A link to the survey will be placed on our departmental webpages hosted by the University of Warwick as well as the Sibs, UK website. The Qualtrics link will also be shared on social media to further recruit participants (Twitter and Facebook).

Recruitment will start from September 2019 and will continue for up to one year (i.e. until October 2020). We will not be collecting personal information that is identifiable. Nonetheless, the raw data set will only be accessed by the research team with only aggregated data disseminated and shared externally, including with our collaborator, the UK charity Sibs. The group being recruited, adult siblings of people with IDD, are not considered a vulnerable group. The information leaflet and consent form (appendix 1 and 2) will be on the first page of the Qualtrics survey, which participants will need to confirm before they can start the survey (appendix 5).

SECTION 7: INFORMED CONSENT

7.1 Please detail the process for obtaining informed consent.

*Informed consent **must** be obtained prior to the participant undergoing any research activities that are specifically for the purposes of the study. This should involve discussion with potential participants or their legally acceptable representative; the presentation of written materials e.g. participant information leaflet(s) –PIL(s) and consent form, and the opportunity to ask questions.*

To include:

- *How and when informed consent will be obtained- written, verbal etc. provide details and justification. Justification must also be provided if informed consent will **not** be sought or if consent will be assumed (please note this needs to be appropriate to the study type).*
- *Who will be taking consent? What training has been undertaken for this?*
- *When and how potential participants will be issued with the information leaflet, in what format and how long they will be given to consider taking part?*
- *Does the study involve children- if so, will consent be obtained from parents, if not provide clear justification why not.*
- *Are the informed consent materials appropriate for the target audience- consider age / language / literacy levels / cultures etc.*

Informed consent will be obtained via the online survey Qualtrics before participants are able to continue to the survey. Before they consent to specific items and click to consent (consent form – appendix 2) and continue to the survey, they will be provided with the full information (information leaflet - see appendix 1). Participants will be able to save the link and come back to the survey at a later date if they want to take time to consider if they want to take part after they have read the information sheet. A telephone number and email address has been provided for the participants to contact the research team if they have any questions. This is sufficient for informed consent as the participants are adults without intellectual disabilities and therefore are not a vulnerable population.

7.2 Please detail how participants withdraw from the study if they have requested to do so.

The process by which an individual can withdraw their participation from the study without giving a reason or experiencing any detrimental effects e.g. should they not wish to continue with their participation in an interview or focus group.

To include:

- *Consideration for any data already collected up until this point- whether it is possible for this to be removed. E.g. it may not be possible to identify data once submitted for an anonymous survey. This needs to be clear in the participant information leaflet (PIL).*
- *Researchers should specify up to what point participants can withdraw their data from a study and **how** a participant would request this- this also needs to be clear in the participant information leaflet (PIL).*
- *Consideration should be given to when data will be anonymised, analysed, published etc. make sure it is possible/feasible for data to be withdrawn if this is being offered to participants. It may be appropriate to provide a time frame for withdrawal.*

Participants can stop taking part in the survey at any time with no negative consequences if they choose to do so. All they have to do to stop taking part in the researcher is to close the internet tab or browser with the

survey in it. However, as we are collecting no identifying information, participants will not be able to withdraw from the study after they have entered their responses, as we will not be able to identify which data is theirs. This has been made clear to participants on the information sheet (appendix 1).

SECTION 8: DATA COLLECTION, USE & STORAGE (DPA 2018 & GDPR)

For projects involving processing of personally identifiable data, please map the data flow to indicate the data controller(s) and data processor(s). This can be submitted as a separate document if necessary, please see accompanying guidance note from the Information Data Compliance Team.

8.1 Does the project involve the collection, analysis or storage of **personally identifiable data**? No

*'Personal data' is any information relating to an **identified** or identifiable natural person- a 'data subject'.*

*An identifiable natural person is one who can be identified, **directly or indirectly**, in particular by reference to an identifier (such as a name, an identification number, location data, financial data, opinion, an online identifier), or to one or more factors specific to the **physical, physiological, genetic, mental, socio-economic, cultural, race, religion, trade union membership, political beliefs, medical, gender or social identity** of that natural person.*

If yes, please provide details of what will be collected: Although some sensitive data is collected on disability, mental health, socio-economic, ethnic, religion and gender identities of the participants, we are not collecting identifying data as well, so these identity characteristics will remain anonymous throughout. We will be collecting postcodes so that we can extract data from the census about neighbourhood deprivation.

Individual will not be identifiable through their postcode. Once the deciles of neighbourhood deprivation have been assigned to each case in the data set, the postcodes will be removed and stored in a separate folder and destroyed after 10 years. All files will be on a password protected, encrypted University of Warwick device and only accessible to the University of Warwick team. All personal data will be deleted after 10 years as per the University of Warwick data retention policy. All personal data will be deleted after 10 years as per the University of Warwick data retention policy.

8.2. Does the project involve the collection, analysis or storage of any **personally identifiable special category data** or **criminal offence data**? No

Special category data includes personal data which is by its nature, particularly sensitive in relation to fundamental rights and freedoms of individuals such as: racial or ethnic origin, political opinions, religious or philosophical beliefs, trade union membership, genetic data, biometric data (for the purpose of identifying a natural person), data concerning health or data concerning a natural person's sex life or sexual orientation. This type of data merits specific protection as the context of its processing. Failure to handle this data correctly could result in significant risks to the fundamental rights and freedoms of the individuals.

If yes, please provide details of what will be collected and for what purpose:

What measures are being implemented to reduce or eliminate the risk to these participants' data for the duration of the period that their personal data is collected and stored? Please see accompanying guidance note for more information.

8.3 Does the project involve the collection or analysis of personal data relating to children under 13 or vulnerable groups? No

UK law provides that for data protection purposes an individual aged under 13 years old is considered a child. For the purposes of the GDPR, a child is someone aged under 16 years old, although Member States are able to reduce this age. Please consider Member State law as Parental/Guardian consent will be required for a child participating in the research.

If yes, please provide details of what will be collected:

For what purpose do you need to process the children's or vulnerable person's data?

What measures are being implemented to reduce or eliminate the risk to these participants' data for the duration of the period that their personal data is collected and stored?

<p>8.4 Who will have access to the study data? <i>Include individuals internal and external to the University and what level of access they have to the data e.g. anonymised, pseudonymised, identifiable etc.</i> <i>Please note you will need to hold a University approved data sharing/processing agreement with each third party (external to the University) with whom data is to be shared.</i></p> <p>Only the research team based at the University of Warwick will have access to the raw data. Other students at the University of Warwick studying an MA Education and Psychology may also analyse these data as part of their Masters theses/dissertations. However they will not work with the version of the data with participant postcodes in, as these postcodes will be stored separately to the analysable data sets. Only aggregated data will be shared externally or publically disseminated.</p>
<p>8.5 During the project, will data be hosted on any external platforms or use new technology? Yes <i>e.g. Apps, online survey tools (qualtrics, Bristol online surveys etc.), recruitment tools (Prolific, SONA etc.), cloud hosting tools.</i></p> <p>If yes, please provide details of the system(s) and how they operate: Data will be collected using Qualtrics. This is on the approved list by the HSSREC and the Information Security team at the University of Warwick.</p> <p>Have you contacted Information Security (informationsecurity@warwick.ac.uk) regarding whether these technologies will be required to go through the Information Assurance workbook approval process? https://warwick.ac.uk/services/idc/informationsecurity/faqs/purchasingissues/ No</p> <p>How and when will the data be deleted and who by?</p> <p>Data hosted on the Qualtrics server will be deleted once data collection is complete and once the downloaded data has been checked (i.e. is the coding all correct). This will be within three months of the survey closing. The survey closes by October 2020, therefore these data will be checked/recorded and deleted from Qualtrics by a CEDAR departmental (based at the University of Warwick) administrator by January 2021.</p>
<p>8.6 Will any research activities be audio or video recorded? No <i>This needs to be clear in the participant information leaflet and consent form.</i></p> <p>If yes, please provide details of what will be recorded, how long it will be kept, how it will be stored securely and how it will be deleted:</p>
<p>8.7 Will data be shared with any organisation external to the University for processing? No <i>e.g. external transcription services, external statistics support, archiving etc.</i></p> <p>If yes, please provide details of the sharing arrangements: clarify whether the data shared will be identifiable, the external organisation to which it will be sent and what contracts/arrangements are in place to safeguard the data and ensure the data processors/controllers will comply with data protection requirements:</p>
<p>8.8 Please detail how, where, in what format and for how long the research data will be stored securely, including on back up storage. <i>e.g. hard/electronic copies, locked filing cabinets in researcher's office, encrypted files, password protected devices, Warwick servers. Please also consider consent forms here. These should be stored separately to research data.</i></p> <p>All data will be downloaded and saved on the University of Warwick M drive, in encrypted, password protected files that only the research team will have access to.</p>
<p>8.9 For this project, will data be processed, (to include the collation, collecting, distributing, sharing, accessing, reviewing, amending, deletion) transferred or stored in any Countries outside UK? No <i>e.g. the use of transcribing service outside the UK, market research company, cloud hosting provider</i></p> <p>If yes, please provide details of the country/countries and the collection/transfer/storage arrangements:</p>
<p>8.10 Describe compliance and proportionality measures in place to satisfy the requirements of the Data Protection Act 2018 and the GDPR. <i>e.g. how will you ensure: fairness and transparency to research participants, data quality, data minimisation (only collect data which is necessary for the purpose(s) of the study), purpose limitation (no further processing of the data for purposes incompatible to those for which it was collected), de-identification of the data as soon as possible, appropriate technical and organisational measures in place to avoid unauthorised access and accidental loss or damage to data</i></p>

etc. Please see accompanying guidance note from the Information Data Compliance Team to help answer this question.

The information sheet (appendix 1) makes it clear how the participants' data will be managed and used. No identifying data is being collected in order to minimise risks to our participants' personal data. As stated, we have carefully constructed the survey to only collect the data necessary to answer our research questions. The data will be downloaded from Qualtrics regularly, and saved on the University of Warwick server, where it is regularly backed up, protecting the integrity of the data.

8.11 Is it anticipated that there will be any future use of the data? No

If yes, please provide details (if known at this stage). This should be clear in the Participant Information Leaflet and on the consent form if there is potential for future use of this data:

SECTION 9: DISSEMINATION

Please describe the dissemination arrangements for the study:

To include:

- *What will happen to the results at the end of the study*
- *Will this study have any pathways to impact? ('Pathways to Impact' are activities designed to ensure any potential impact is realised, measured and evidenced.)*
- *How and where will the results be reported/published?*
- *Are there any plans to notify/debrief the participants of the outcome of the study, either by provision of the publication, or via a specifically designed newsletter, presentation etc.?*
- *If it is possible for the participant to specifically request results from the researcher when would this information be provided e.g. after the Final Study Report had been compiled or after the results had been published?*

The results will be compiled into a paper to be submitted for publication in an academic journal. The study will also be disseminated at conferences in poster or oral formats. A brief, summary report and video clips will be developed with the UK charity Sibs for them to disseminate on their website and mailing list. These summaries can also be shared with the '1,000 Families Study' mailing list with a link to the publication. The links with Sibs will provide a pathway to impact. Dissemination activities are being planned as part of an 'Overseas Institutional Visit' next academic year and early results of this study may contribute to that dissemination activity.

SECTION 10: FURTHER INFORMATION (OPTIONAL)

Please provide any further details/information relevant to this application that may aid the ethical review process.

To include:

- *For complex studies with multiple work packages, collaborators or steering groups, applicants may wish to submit a protocol or supplementary documents in addition to this application form detailing the roles and responsibilities of each party.*
- *Projects that require further approvals e.g. HRA approval for research in the NHS may also wish to submit a protocol for review.*
- *Peer review*
- *Patient and public involvement*
- *Flow diagram*
- *Data management plan*

The topics to be covered in this survey have been developed in collaboration with the UK charity Sibs, the staff of which are also adult siblings.

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SECTION 11: SUPPORTING DOCUMENTS

HSSREC will need to review **all** participant facing documents associated with this application.

There may be more than one type of each document for each study, i.e. multiple participant information leaflets if there are different participant groups, or work packages.

Please specify below, which documents have been submitted with this application (where applicable):

- Participant information leaflet(s)
- Consent form(s)
- Poster(s)/advertisement(s)
- Invitation email(s)
- Questionnaire(s)/Survey question(s)
- Interview schedule(s)/topic guide(s)
- Data Collection form
- Data flow map
- Data Management Plan
- Risk assessment
- Protocol (optional- needs to be consistent with the application)
- Other, please specify:

SECTION 12. SIGNATURES AND DECLARATIONS

The information in this form together with any accompanying information is complete and correct to the best of my knowledge and belief and I take full responsibility for it.

I undertake to abide by the University of Warwick's Research Code of Practice in undertaking this study.

I understand that HSSREC grants ethical approval for projects, and that the seeking and obtaining of all other necessary approvals and permissions prior to starting the project is my responsibility.

I confirm I am familiar with and will conduct my project in line with the General Data Protection Regulation (GDPR) and Data Protection Act 2018 (DPA 2018), reporting any data breaches to the University's Information and Data Director: DPO@warwick.ac.uk.

I understand that I must not begin research and related projects with human participants, their data or tissue until I have received full approval from the relevant Research Ethics Committee of the University of Warwick.

I understand that any changes that I would like to make to this study after receiving approval from HSSREC, require further review. As such they must be submitted to hssrec@warwick.ac.uk before such changes are implemented.

Signature of Applicant:

Date:

3.2: Signed Declaration for Ethical Approval

SECTION 11: SUPPORTING DOCUMENTS

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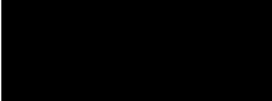
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I understand that any changes that I would like to make to this study after receiving approval from HSSREC, require further review. As such they must be submitted to hssrec@warwick.ac.uk before such changes are implemented.

Signature of Applicant: 25-6-2019



Signature of Supervisor (If applicable):		Date: 25-6-2019
Signature of Head of Department:		Date: 25-6-2019

Note. Your electronic submission should contain signatures (electronic signatures will be accepted) of all relevant parties. Applications without the necessary signatures will be returned

Please send an electronic copy of the application to hssrec@warwick.ac.uk

If you have not already done so, you are strongly recommended to undertake the Research Integrity Online Training Course. All details relating to this course can be found [here](#).

3.3: Participant Information Sheet– Form was Uploaded onto Qualtrics



Participant Information Leaflet version 4 (online final version)

Participant Information Leaflet

Study Title: Adult sibling survey

Investigator(s): Nikita Hayden, Professor Richard Hastings

Introduction

You are invited to take part in a research study. Before you decide, you need to understand why the research is being done and what it would involve for you. Please take the time to read the following information carefully. Talk to others about the study if you wish.

Please ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

Who is organising and funding the study?

A PhD researcher, under supervision, funded by the Economic and Social Research Council and the UK charity Sibs is conducting this study. Sibs have contributed to the design of this study.

What is the study about?

The study seeks to further understand the experiences, sibling relationships, wellbeing and the needs of adults who have a brother or sister with a learning or developmental disability. By learning/developmental disability, we mean: Autism, Cerebral palsy, Fragile X syndrome, Down syndrome, genetic disorders etc. Your brother or sister may have one of these conditions, or several. We will refer to these conditions collectively as learning/developmental disabilities throughout this study. If you agree, you will complete an online survey about these topics. We also want to explore adult siblings' caregiving role or their expectations for caring in the future, as well as the support needs of siblings of people with intellectual/developmental disabilities. This is important, because it will help us consider ways to support the wellbeing of siblings of people with intellectual/developmental disabilities and to support whatever role they decide to take in the lives of their brothers and sisters.

Who is this study for?

This study is for adults (aged 18 years and over) living in the UK that have a brother or a sister with learning/developmental disability. This might be an intellectual disability, autism, Down syndrome or Fragile X syndrome, Cerebral Palsy, amongst other conditions.

What would taking part involve?

If you decide to take part in the study, once you have read to the end of this information page, you should then consent (if you agree) to the conditions at the bottom of this webpage. You will then be able to progress to the survey. In this survey, you will be asked questions about you, your socio-demographic background, your wellbeing, your brother or sister with a

learning/developmental disability and your relationship with them, what care you give (if any) and what support you need. The survey takes about 20 to 25 minutes to complete. This will all be anonymous because we will not ask you for your name or contact information.

Do I have to take part?

No. Participation in this study is completely voluntary and choosing not to take part will not affect you or your family in any way. All you need to do is close the webpage to stop taking part in this study. Further details about withdrawing from the study are provided later on this webpage.

What are the possible benefits of taking part in this study?

Taking part in this research gives you an opportunity to reflect on your experiences and contribute to our understanding about the experiences and needs of adult siblings of people with learning/developmental disabilities. This research is being conducted in collaboration with Sibs, therefore the findings will inform their priorities in practice.

What are the possible disadvantages, side effects or risks, of taking part in this study?

We do not anticipate any risks to the participants of this study or their families. We will not be asking for your name, to protect your anonymity. Most questions that we are asking have been used in several research studies before. However, it is possible that you will find some of the questions to be upsetting because we do ask about your wellbeing and some of the challenges faced by you and your brother/sister with learning/developmental disability. If you are upset by any of the questions, you do not have to respond to them and you are under no obligation to continue with the survey.

If any of the survey or interview questions make you concerned for yourself or another family member's wellbeing, we recommend that you make contact with your General Practitioner (GP) or by requesting a call back from Sibs: <https://www.sibs.org.uk/contact/>, or by calling the Mencap helpline: 0808 808 1111.

Expenses and payments

There are no expenses or payments being offered as part of this study.

Will my taking part be kept confidential?

Yes, all information about you will be handled in confidence and all information will be kept securely (on secure, password protected, encrypted computers) in an anonymised form. Information from the study will only be seen by the research team. Your data will be collected via an online survey (Qualtrics). The survey asks you to provide your postcode. We would like this information because we can then get other information from the UK Census about the area in which you live. You do not have to provide this information. Postcodes will be kept separate from all other data and protected on an encrypted University of Warwick server.

Only members of the research team will have access to data. However, in some instances, officials from regulatory authorities may need to access data for checking the quality of the research. All members of the research team and regulatory bodies are trained in data protection issues. Study information will be kept securely for up to 10 years in line with the University of Warwick's policies. No personal data will be collected therefore it will not be shared with anyone or shared outside of the EEA. We would like to publish direct quotes provided by you and you will be asked to consent to this at the bottom of this webpage.

What will happen to the data collected about me?

As a publicly-funded organisation, the University of Warwick have to ensure that it is in the public interest when we use personally-identifiable information from people who have agreed

to take part in research. This means that when you agree to take part in a research study, such as this, we will use your data in the ways needed to conduct and analyse the research study.

We will be using information from you in order to undertake this study and will act as the data controller for this study. We are committed to protecting the rights of individuals in line with data protection legislation. The University of Warwick will keep your personal responses for 10 years after the end of the study after which, these data shall be deleted. The collective responses of everyone that completes the survey will be written up in a report and an academic paper and will be made publically available indefinitely.

No identifiable data will be collected from you as part of this study. This means that once your responses have been submitted to the research team, it will not be possible to withdraw this data as your individual responses cannot be identified.

What will happen if I don't want to carry on being part of the study?

Your participation in this study is completely voluntary. You are free to stop participating in the survey whenever you wish to, and this will not negatively effect you in any way. You do not need to answer any questions that you are uncomfortable answering. If you withdraw from the study it will not be possible to withdraw your data that has already been collected as your responses are all anonymised, as we will not collect any personal identifying information from you in the survey.

What will happen to the results of the study?

The results will be compiled into a paper to be submitted for publication in an academic journal. The study will also be disseminated at conferences in poster or oral formats. A brief, 'easy-read' summary report will be shared with relevant third sector organisations, including the UK charity Sibs. Dissemination activities are being planned as part of an 'Overseas Institutional Visit' next academic year and the early results of this study may contribute to that dissemination activity.

Who has reviewed the study?

This study has been reviewed and given favourable opinion by the University of Warwick's Humanities and Social Science Research Ethics Committee. Reference number HSSREC 137/18-19, date 29-07/2019.

Who should I contact if I want further information?

Nikita Hayden, PhD student, Centre for Educational Development, Appraisal and Research, University of Warwick.



Nikita Hayden's supervisor:

Professor Richard Hastings, Professor and Cerebra Chair of Family Research, Centre for Educational Development, Appraisal and Research, University of Warwick.



Who should I contact if I wish to make a complaint?

Any complaint about the way you have been dealt with during the study or any possible harm you might have suffered will be addressed. Please address your complaint to the person below, who is a senior University of Warwick official entirely independent of this study:

Head of Research Governance
Research & Impact Services

University House
University of Warwick
Coventry
CV4 8UW
Email: researchgovernance@warwick.ac.uk
Tel: 024 76 522746

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer, Anjeli Bajaj, Information and Data Director who will investigate the matter: DPO@warwick.ac.uk.

If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Thank you for taking the time to read this Participant Information Leaflet

3.4: Participant Consent Form – Form was Uploaded onto Qualtrics



Note this information will be presented as a part of an online survey.

Title of Project: *Adult sibling survey*

Name of Researcher(s): *Nikita Hayden, Professor Richard Hastings.*

*Please read the statements below carefully. If you agree with these statements then tick in the corresponding box. **Unfortunately, if you do not consent to all the statements we cannot use your survey responses.***

Please initial all boxes

1. I confirm that I have read and understand the information sheet for the above study. I have asked any questions I wanted to ask.
2. I confirm that I am aged 18 years or older.
3. I confirm that I have a brother or sister with a learning (intellectual)/developmental disability.
4. I confirm that I reside in the UK.
5. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason.
6. I understand that data collected during the study may be looked at by individuals from The University of Warwick, from regulatory authorities where it is relevant to my taking part in this study. I give permission for these individuals to have access to my data.
7. I understand that my data will be securely stored for a minimum of 10 years, in line with the University of Warwick's Research Data Management Policy.
8. I consent to the use (anonymised) of my verbatim quotations.
9. I am happy for my data to be used in future research.
10. I agree to take part in the above study.

3.5: Adult Sibling Survey – Survey was Uploaded onto Qualtrics

Adult Sibling survey FINAL ONLINE VERSION

1

Adult sibling survey – Adults who have a brother or sister with a learning or developmental disability (e.g. Autism, Cerebral palsy, Fragile X syndrome etc.)

The study will ask you to answer questions about yourself and your brother or sister with a learning or developmental disability. By learning/developmental disability, we mean: Autism, Cerebral palsy, Fragile X syndrome, Down syndrome, genetic disorders etc. Your brother or sister may have one of these conditions, or several. We will refer to these various disabilities under the umbrella term 'learning/developmental disability' for the remainder of this survey. If you have more than one sibling with a learning/developmental disability, please select your nearest-in-age sibling with a learning/developmental disability and answer these questions about them throughout this survey.

Questions about you

Your age (In years)	
----------------------------	--

Your gender (please select one)	✓
Male	
Female	
Other (please describe below)	

Questions about your ethnicity and religion

In this section, you will be asked to indicate which category best describes your ethnicity and religion. The reason we are asking you these questions is that not much is known about how things such as ethnicity and religion impact on experiences of having a family member with learning/developmental disability. Some siblings have told us that they think these cultural factors may be important factors for us to consider so we would like to be able to explore these cultural differences in this study. You may skip these questions if they make you feel uncomfortable.

How would you describe your ethnicity?¹

Please select ONE	✓
Asian/Asian British: Indian	
Asian/Asian British: Pakistani	
Asian/Asian British: Bangladeshi	
Asian/Asian British: Chinese	

¹ Census forms (2011). 2011 England – Household, 2011 Census form. Office for National Statistics, UK Data Service website. Retrieved from <https://census.ukdataservice.ac.uk/use-data/censuses/forms.aspx>. Retrieved 19th June 2019.

Adult Sibling survey FINAL ONLINE VERSION

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Asian other (please describe below)	

Black/African/Black British: African	
Black/African/Black British: Caribbean	
Black other (please describe below)	

Mixed/multiple ethnic groups: White and Black Caribbean	
Mixed/multiple ethnic groups: White and Black African	
Mixed/multiple ethnic groups: White and Asian	
Mixed other (please describe below)	

Other Ethnic group: Arab	
Ethnic other (please describe)	

White: English/Welsh/Scottish/Northern Irish/British	
White: Irish	
White: Travelling community	
White: Other (Please describe below)	

Any other ethnic background (Please describe below)	

How would you describe your religion?² <i>Please select one</i>	✓
No religion	
Christian (<i>including Church of England, Catholic, Protestant and all other Christian denominations</i>)	
Buddhist	
Hindu	
Jewish	
Muslim	
Sikh	

² *Ibid.*

Adult Sibling survey FINAL ONLINE VERSION

3

Any other religion (please describe below)	

What is your country of residence? <i>Please select one</i>	✓
England	
Northern Ireland	
Scotland	
Wales	
Other (please describe below)	

Your postcode

We would like this information to be able to access UK national data based on the 2011 Census about the neighbourhood in which you live. If your postcode is not accepted, please type it instead in the final free text box (question 41) – along with your response to that question.

--

Questions about your household

*Data from research with families with a family member with a learning/developmental disability has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether people with different levels of financial resources have different experiences. **Please remember if any of these questions make you uncomfortable, then you can skip them without answering and move on to the next set of questions. This survey is all fully anonymous, therefore you will not be identified in any way.***

What is your total weekly household income (after any deductions e.g. income tax), including income from paid work, pension, Social Services Benefits (e.g. Job Seekers Allowance, DLA, Carers' Allowance, Attendance Allowance, Tax Credits, Housing Benefits, Pension Credits) etc.?

Please select <u>ONE</u>	✓
£200 or less	
Between £201 and £300	
Between £301 and £400	
Between £401 and £500	
Between £501 and £600	

Adult Sibling survey FINAL ONLINE VERSION

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Between £601 and £700	
Between £701 and £800	
Between £801 and £900	
Between £901 and £1000	
Over £1000	

How well would you say you [and your husband/wife/partner] are managing financially these days? Would you say you are ...

Please select ONE	✓
living comfortably?	
doing alright?	
just about getting by?	
finding it quite difficult?	
finding it very difficult?	

Suppose you only had one week to raise £2000 for an emergency, which of the following best describes how hard it would be for you to get that money?

Please select ONE	✓
I could easily raise the money	
I could raise the money, but it would involve some sacrifices (e.g. reduced spending, selling a possession)	
I would have to do something drastic to raise the money (e.g. selling an important possession)	
I don't think I could raise the money	

In total how many people currently live in your home (including yourself)?

	Individuals over the age of 14	Individuals under the age of 14
Number		

Do you currently live in your parental home?

Please select ONE	✓
No	
Yes	

Do you currently live with your sibling(s) with learning/developmental disability?

Please select ONE	✓
No	
Yes	

Please select the highest level of your educational qualifications

Please select ONE	✓
No qualifications	

Adult Sibling survey FINAL ONLINE VERSION

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Some GCSEs passes or equivalent	
5 or more GCSEs at A*-C or equivalent	
A/AS Levels or equivalent	
Higher Education but below degree level (i.e. foundation degree diploma , higher apprenticeship, Level 4 and Level 5 qualifications)	
Degree (e.g. BA, BSc, MA)	
Don't know	

What is your current work status?

Please select <u>ONE</u>	✓
Employed or self-employed full time (30+ hours/week)	
Employed or self-employed part-time (or unknown hours)	
Employed or self-employed but on long-term leave (e.g. maternity, paternity, sick)	
Full-time carer	
In education/student	
Not working and looking for work	
Not working and not looking for work	

What is your current marital status?

Please select <u>ONE</u>	✓
Single	
Married/civil partnership and living with spouse/civil partner	
Living with partner	
Divorced /Separated /Widowed /Not currently living with partner	

Your health and wellbeing**How is your health in general?³**

Please select <u>ONE</u>	✓
Very good	
Good	
Fair	
Bad	
Very bad	

Do you have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled you over a period of time or is likely to affect you over a period of time?

Please select <u>ONE</u>	✓
No	
Yes	

³ Survey of Carers in Households (2010). 451845 - Survey of Informal Carers in Households 2009/10 Mainstage questionnaire. *Survey of Carers in Households – England, 2009-10*. NHS digital, National statistics. Retrieved from <https://files.digital.nhs.uk/publicationimport/pub02xxx/pub02200/surv-care-house-care-ques-eng-2009-2010-apx2.pdf>. Retrieved 19th June 2019.

Adult Sibling survey FINAL ONLINE VERSION

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Below are some statements about feelings and thoughts. Please select the answer that best describes your experience of each over the last 2 weeks.

Please select ONE answer per statement

Statements	None of the time	Rarely	Some of the time	Often	All of the time
I've been feeling optimistic about the future					
I've been feeling useful					
I've been feeling relaxed					
I've been dealing with problems well					
I've been thinking clearly					
I've been feeling close to other people					
I've been able to make up my own mind about things					

This measure is the Short Warwick Edinburgh Well-Being Scale (SWEMWBS, 2019)⁴ © NHS Health Scotland, University of Warwick and University of Edinburgh, 2007, all rights reserved.

For each question, please select the answer that best describes how often you had this feeling.

During the past 30 days, about how often do you feel-

Please select ONE answer per statement

Please select <u>ONE</u> answer per statement	All of the time	Most of the time	Some of the time	A little of the time	None of the time
...nervous?					
...hopeless?					
...restless or fidgety?					
...so depressed that nothing could cheer you up?					
...that everything was an effort?					
...worthless?					

This measure is the Kessler Psychological Distress Scale (K6; Kessler, et al., 2003).⁵

If we were to define 'quality of life' as how you feel overall about your life, including your standard of living, your surroundings, friendships and how you feel day-to-day, how would you rate your quality of life? ⁶	✓
Very good	
Fairly good	

⁴ WEMWBS (2019). Warwick-Edinburgh Mental Wellbeing Scale – (S) WEMWBS. *Warwick Medical School, University of Warwick*. Retrieved from <https://warwick.ac.uk/fac/sci/med/research/platform/wemwbs/>. Retrieved 9th March 2019.

⁵ Kessler, R.C., et al. (2003). Screening for serious mental health difficulties in the general population. *Archives of General Psychiatry*. 60, 184-189.

⁶ Survey of Carers in Households (2010). *Op cit*.

Adult Sibling survey FINAL ONLINE VERSION

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Neither good nor bad	
Fairly bad	
Very bad	

About your brother or sister with a learning/developmental disability

In this section we would like you tell us about your sibling with a learning/developmental disability. Remember, if you have more than one sibling with a learning/developmental disability, please select your nearest-in-age sibling with a learning/developmental disability and answer these questions about them throughout.

How many siblings do you have (both with and without disabilities)?	
--	--

How many of these siblings have a disability?	
--	--

How old is your sibling with a learning/developmental disability? (In years)	
---	--

What gender is your sibling with a learning/developmental disability? (please select one)	✓
Male	
Female	
Other (please describe below)	

Please select which of the conditions below have been used to describe your brother or sister with learning/developmental disability: <i>Please select all that apply</i>	✓
Learning disability/learning difficulties/intellectual disability	
Autism/Autistic Spectrum Disorder/Autistic Spectrum Condition/Asperger's Syndrome	
Down syndrome	
Fragile X Syndrome	
Cerebral palsy	
Other genetic syndrome/diagnosis <i>Please specify</i>	

Does your brother or sister with learning/developmental disability also have a:
--

Adult Sibling survey FINAL ONLINE VERSION

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	Yes	No	I do not know
Visual impairment?			
Hearing impairment?			
Mobility problems			
Physical health problems			
Epilepsy/seizures			

We would like to know about your brother or sister's current level of independence in performing activities of daily living. For each activity please tell us the number which best describes your brother/sister's ability to do the task. For example, independent would mean your brother or sister is able to do the task without any help or assistance. Please select ONE answer per statement

Please rate your brother or sister's level of independence in:	Ability to perform task			
	Independent or does on own	Does with help	Does not do at all	I do not know
Making their own bed				
Doing household tasks, including picking up around the house, putting things away, light housecleaning, etc.				
Doing errands, including shopping in stores				
Doing home repairs, including simple repairs around the house, non-technical in nature; for example, changing light bulbs or repairing a loose screw				
Doing laundry, washing and drying				
Washing/bathing				
Grooming, brushing teeth, combing and/or brushing hair				
Dressing and undressing				
Toileting				
Preparing simple foods requiring no mixing or cooking, including sandwiches, cold cereal, etc.				
Mixing and cooking simple foods, fry eggs, make pancakes, heat food in microwavc, etc.				
Preparing complete meal				
Setting and clearing table				
Drinking from a cup				
Eating from a plate				
Washing dishes (including using a dishwasher)				
Banking and managing daily finances, including keeping track of cash, checking account, paying bills, etc. (Note: if he/she can do a				

Adult Sibling survey FINAL ONLINE VERSION

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portion but not all indicate “does with help”				
---	--	--	--	--

This measure of intellectual (learning) disability support needs is the Waisman Activities of Daily Living Scale (W-ADL; Maenner, 2013).⁷

Who does your brother or sister with learning/developmental disability live with?	✓
Parents	
Another family member	
Alone	
In a group home	
With friends	
With a romantic partner	
You	
In supported living	
Other, please specify:	

What tools, resources and information do you currently have to help you support your brother or sister with learning/developmental disability? <i>Please elect all that apply</i>	✓
I have no tools, resources or information to help me	
Friends and family	
Another/other siblings also supporting brothers or sisters with a learning/developmental disability	
A formal group that supports siblings of people with learning/developmental disabilities	
Facebook groups	
Books	
Counselling or other psychological support	
Other online groups	
Sibs charity	
Disability support/advocacy charities	
Other (<i>please describe below</i>)	

⁷ Maenner MJ, Smith LE, Hong J, Makuch R, Greenberg J, Mailick MR. (2013). An Evaluation of an Activities of Daily Living Scale for Adolescents and Adults with Developmental Disabilities. *Disability and Health Journal*, 6(1), 8-17. doi:10.1016/j.dhjo.2012.08.005

Caring and support for your brother or sister with learning/developmental disability

Do you consider yourself a caregiver for your brother/sister with learning/developmental disability? <i>A carer is anyone, including children and adults, who looks after a family member, partner or friend who needs help because of their illness, frailty, disability, a mental health problem or an addiction and cannot cope without their support. The care they give is unpaid. Please select one</i>	✓
No	
Yes (please describe below)	

Which role are you likely to fulfil (or have already taken on) for your brother/sister with a learning/developmental disability when your parents are no longer able to provide care? <i>A secondary or shared caregiving role means that you share caregiving with more than one person.</i>			
	No caregiving role	Secondary/shared caregiving role	Primary caregiver
Anticipated future role			

If you do provide a supporting role for your brother or sister with a learning/developmental disability, rate the intensity of support you provide on a scale from 1 to 10 (1 being none and 10 being full)	
---	--

Has your ability to spend time doing leisure or social activities been affected by the assistance you give to your sibling with a learning/developmental disability in any of the ways described below? (For all responses, this should be as a result of the caring and not for other reasons). Please select <u>ANY</u> that apply.	✓
Unable to socialise or take part in social or leisure activities at all (due to caring responsibilities)	
Reduced time with spouse or partner	
Reduced time with other family members	
Reduced time with friends	
Difficulties making new friends	
Reduced time spent doing sport or physical activity	
Reduced time spent doing pastime or hobby	
None of these	
Other. Please specify	

This measure of carer burden is from the Survey of Carers in Households (2010).⁸

⁸ Survey of Carers in Households (2010). *Op cit.*

Adult Sibling survey FINAL ONLINE VERSION

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Your relationship with your brother or sister with learning/developmental disability

Please tell us about your relationship with your brother or sister with a learning/developmental disability by selecting one response to each statement below.					
Statement	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree
1. My sibling makes me happy.					
2. My sibling's feelings are very important to me.					
3. I enjoy my relationship with my sibling.					
4. I am proud of my sibling.					
5. My sibling and I have a lot of fun together.					
6. My sibling frequently makes me very angry.					
7. I admire my sibling.					
8. I like to spend time with my sibling.					
9. I presently spend a lot of time with my sibling.					
10. I call my sibling on the telephone frequently.					
11. My sibling and I share secrets.					
12. My sibling and I do a lot of things together.					
13.* I never talk about my problems with my sibling.					
14. My sibling and I borrow things from each other.					
15. My sibling and I 'hang out' together.					
16. My sibling talks to me about personal problems.					
17. My sibling is a good friend.					
18. My sibling is very important in my life.					

Adult Sibling survey FINAL ONLINE VERSION

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19. My sibling and I are not very close.					
20. My sibling is one of my best friends.					
21. My sibling and I have a lot in common.					
22. I believe I am very important to my sibling.					
23. I know that I am one of my sibling's best friends.					
24. My sibling is proud of me.					

This sibling relationship measure is a reduced version of the Lifespan Sibling Relationship Questionnaire (LSRS; Riggio, 2000).⁹

Your needs in relation to your brother or sister with learning/developmental disability

This section asks you questions about your needs in terms of information and support in relation to your brother or sister with learning/developmental disability. Please read each statement listed below and indicate how important it would be for you to have support in each of the areas.

		I definitely do not need help with this	Not sure	I definitely need help with this
Needs for Information				
1	I need more information about my sibling's learning/developmental disability			
2	I need more information about how to handle my sibling's behaviour			
3	I need more information about how to teach my sibling			
4	I need more information on how to spend time with or talk to my sibling			
5	I need more information on the services that are presently available for my sibling			
6	I need more information about the services that my sibling might receive in the future			
7	I need more information about how people with learning/developmental disability grow and develop			
Needs for Support				
8	I need to have someone in my family that I can talk to more about problems			

⁹ Riggio, H. R. (2000). Measuring attitudes toward adult sibling relationships: The Lifespan Sibling Relationship Scale. *Journal of Social and Personal Relationships*, 17(6), 707-278.

Adult Sibling survey FINAL ONLINE VERSION

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9	I need to have more friends that I can talk to			
10	I need to have more opportunities to meet and talk with other siblings of people with learning or developmental disabilities like myself			
11	I need to have more time just to talk with my sibling's carers and support workers			
12	I would like to meet more regularly with a counsellor (psychologist, social worker, psychiatrist) to talk about problems			
13	I need to talk more to a Vicar, Imam, Priest, Rabbi, or other religious leader who could help me deal with problems			
14	I need reading material about other siblings who have a brother or sister similar to mine			
15	I need to have more time for myself			
Explaining to Others				
16	I need more help in how to explain my sibling's condition to other people in our family			
17	I need more help in explaining my sibling's condition to either my spouse or my spouse's parents			
18	My spouse needs help in understanding and accepting my sibling's condition			
19	I need help in knowing how to respond when friends, neighbours, or strangers ask questions about my sibling's condition			
20	I need help in explaining my sibling's condition to other people			
Community Services				
21	I need help in getting appropriate acceptance for my sibling in our church, synagogue, Mosque, Gurdwara or other place of worship during religious services			
Financial needs				
22	I need more help in paying for expenses such as food, housing, medical care, clothing, or transportation			
23	I or my spouse need more counselling or help in getting a job			
Family Functioning				
24	Our family needs help in discussing problems and reaching solutions			
25	Our family needs help in learning how to support each other during difficult times			
26	Our family needs help in deciding who will do household chores, sibling care, and other family tasks			

27	Our family needs help deciding on and doing recreational activities			
----	---	--	--	--

This Family Needs measure has been reduced and adapted from Bailey & Simeonsson (1988)¹⁰ for adult siblings.

Your views of being a sibling of someone with a learning/developmental disability

Is there anything else you want to tell us about being a sibling of someone with a learning/developmental disability? *For example you might want to tell us more about your relationship with your brother or sister with learning/developmental disability, your thoughts on your brother or sister with learning/developmental disability, or your experiences as a sibling of someone with a learning/developmental disability.*

¹⁰ Bailey, D. B. & Simeonsson, R. J. (1988). Assessing Needs of Families with Handicapped Infants. *The Journal of Special Education*. 22, 1. 117-127.

3.6: Study Poster and Email Invitation



DO YOU HAVE A BROTHER OR SISTER WITH A LEARNING (INTELLECTUAL)/DEVELOPMENTAL DISABILITY (E.G. AUTISM)?

Would you be interested in telling us about your experiences?

We are looking for adults (aged 18 years or older), living in the UK that have a brother or sister with an learning/developmental disability to take part in a brief online survey.

Your brother or sister's learning/developmental disability might include Autism, intellectual disability, Down syndrome, Cerebral palsy, Fragile X syndrome, genetic disorders, etc.

To find out more and take part:

Follow this link [XXXXXXXXXXXXXXXXXXXXXXXXXX](#).

If you have any questions, please contact
Nikita Hayden

Email: [\[REDACTED\]](#)

Invitation to take part in the study email

This email will be sent to local intellectual/learning/developmental disability support charities organisations and Sibs to ask them to send this invite to their mailing list to recruit adult siblings to take part in the study.

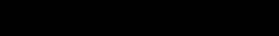
Dear XX,

We are looking for adults that have a brother or sister with a learning (intellectual)/developmental disability to take part in our survey study. We want to hear about your experiences, wellbeing, your relationship with your brother or sister with a learning/developmental disability, as well as what your support needs are. Developmental disabilities may include Autism, Cerebral palsy, Fragile X syndrome, Down syndrome, genetic disorders etc. Please see the attached poster with further information about the study (appendix 3). This research is collaborative with the UK charity Sibs

To take part, you need to be 18 years of age or older, have a brother or sister with a learning/developmental disability and reside in the UK.

This research is important because understanding the social, psychological and health outcomes of adult siblings will help us develop relevant support to improve outcomes and relationships between siblings where one has a learning/developmental disability. As this work is collaborative with the UK charity Sibs, this is a great opportunity to have your needs and experiences heard by the only UK charity whose sole purpose is to support siblings of people with disabilities.

If you are interested in finding out more about the study, please follow this link to the survey XX. If you have any questions about this study, you can contact Nikita Hayden via



Best wishes,

Nikita Hayden

PhD student

University of Warwick

3.7: Conditional Approval Letter from Ethics Committee



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Humanities and Social Sciences Research Ethics Committee
Kirby Corner Road
Coventry
CV4 8UW

Wednesday, 17 July 2019

Ms Nikita Hayden
CEDAR
University of Warwick
Coventry
CV4 7AL

Dear Ms Hayden,

Ethical Application Reference: HSSREC 137/18-19
Title: Adult Sibling Survey

Thank you for submitting your ethics application to the Humanities and Social Sciences Research Ethics Committee (HSSREC) for consideration. Your application has been reviewed and **conditional approval** has been granted. This means that queries about aspects of the study and the associated documentation have been raised, these are detailed below. **These must be addressed and reviewed by HSSREC before full approval can be given.**

1. Throughout the application it is assumed that the sibling will not have a disability of their own, please clarify whether this is an inclusion/exclusion criteria and what levels of disability would be included/excluded. For example, if the sibling has underlying mental health issues, would they be excluded on these grounds?
2. On the Adult Sibling Survey p. 7, please change the question 'Making his/her own bed' so that it is not gender binary.

How to submit responses:

Please amend the original application form and other relevant documentation, with changes clearly highlighted, and include in your resubmission a word document summarising the changes you have made.

Your revised documents also need to be distinguishable as different versions to those originally submitted. The recommended method for doing this is to include the appropriate version numbers and dates of submission in the footer.

When completed, please email your revised documents to hssrec@warwick.ac.uk



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Your revised application will be reviewed by HSSREC and you can expect a response within 10 working days.

Yours sincerely



Dr Fiona MacCallum
Chair, Humanities and Social Sciences Research Ethics Committee

3.8: Approval Letter from Ethics Committee



WARWICK
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Humanities and Social Sciences Research Ethics Committee
Kirby Corner Road
Coventry
CV4 8UW

Monday, 29 July 2019

Ms Nikita Hayden
CEDAR
University of Warwick
Coventry
CV4 7AL

Dear Ms Hayden,

Ethical Application Reference: HSSREC 137/18-19

Title: Adult Sibling Survey

Thank you for submitting your revisions to the Humanities and Social Sciences Research Ethics Committee (HSSREC) for consideration. We are pleased to advise you that, under the authority delegated to us by the University of Warwick Research Governance and Ethics Committee, **full approval for your project is hereby granted.**

Before conducting your research it is strongly recommended that you complete the on-line Research Integrity training:

www.warwick.ac.uk/ritraining. Support is available from the HSSREC Secretary.

In undertaking your study, you are required to comply with the University of Warwick's Research Code of Practice:

https://warwick.ac.uk/services/ris/research_integrity/code_of_practice_and_policies/research_code_of_practice/

You are also required to familiarise yourself with the University of Warwick's Code of Practice for the Investigation of Research Misconduct:

https://warwick.ac.uk/services/ris/research_integrity/research_misconduct/codeofpractice_researchmisconduct/

You must ensure that you are compliant with all necessary data protection regulations:

<https://warwick.ac.uk/services/idc>

Please ensure that evidence of all necessary local permissions is provided to HSSREC prior to commencing your study.

Please also be aware that HSSREC grants **ethical approval** for studies. The seeking and obtaining of all other necessary approvals is the responsibility of the investigator.

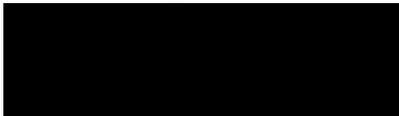


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Any substantial changes to any aspect of the project will require further review by the Committee and the PI is required to notify the Committee as early as possible should they wish to make any such changes. The HSSREC Secretary should be notified of any minor amendments to the study.

May I take this opportunity to wish you the very best of luck with this study.

Yours sincerely



Dr Fiona MacCallum
Chair, Humanities and Social Sciences Research Ethics Committee

Appendix Four: Ethics and Study Documentation for Chapter Five, Study Four

4.1: Application for Ethical Approval – Final Version

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

Please enter a short title for this project (maximum 70 characters)

The 1,000 Families Study

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located?(Tick all that apply)

- England
- Scotland
- Wales
- Northern Ireland

NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

3a. In which country of the UK will the lead NHS R&D office be located:

- England
 Scotland
 Wales
 Northern Ireland
 This study does not involve the NHS

4. Which review bodies are you applying to?

- NHS/HSC Research and Development offices
 Social Care Research Ethics Committee
 Research Ethics Committee
 Confidentiality Advisory Group (CAG)
 National Offender Management Service (NOMS) (Prisons & Probation)

For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.

5. Will any research sites in this study be NHS organisations?

- Yes No

6. Do you plan to include any participants who are children?

- Yes No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- Yes No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

- Yes No

9. Is the study or any part of it being undertaken as an educational project?

- Yes No

Please describe briefly the involvement of the student(s):
Three PhD students will work on data collection for this survey. Each will be examining and reporting data from a different area of the survey, with their own research questions, to contribute towards their PhD theses.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

- Yes No

Date: 14/07/2015

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10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

Yes No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

Yes No

DRAFT

Integrated Research Application System
Application Form for Research administering questionnaires/interviews for quantitative analysis or mixed methodology study



Application to NHS/HSC Research Ethics Committee

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
 The 1,000 Families Study

Please complete these details after you have booked the REC application for review.

REC Name:
 West Midlands - South Birmingham

REC Reference Number:
 15/WM/0267

Submission date:
 14/07/2015

PART A: Core study information

1. ADMINISTRATIVE DETAILS

A1. Full title of the research:

The 1,000 families study: Well-being in families of children with intellectual disability

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title	Forename/Initials	Surname
	Miss	Emma	Langley
Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
Post Code	CV47AL		
E-mail	[REDACTED]		
Telephone	[REDACTED]		
Fax	[REDACTED]		

NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:
PhD, Education and PsychologyName of educational establishment:
University of Warwick**Student 2**

	Title	Forename/Initials	Surname
	Miss	Jane	Margetson
Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
Post Code	CV47AL		
E-mail	[REDACTED]		
Telephone	[REDACTED]		
Fax			

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:
PhD, Education and PsychologyName of educational establishment:
University of Warwick**Student 3**

	Title	Forename/Initials	Surname
	Miss	Mikeda	Jess
Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
Post Code	CV47AL		
E-mail	[REDACTED]		
Telephone	[REDACTED]		
Fax			

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:
PhD, Education and PsychologyName of educational establishment:
University of Warwick

Name and contact details of academic supervisor(s):

Academic supervisor 1

	Title	Forename/Initials	Surname
	Professor	Richard	Hastings
Address	CEDAR (Centre for Educational Development Appraisal and Research)		

Date: 14/07/2015

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NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

	University of Warwick Coventry UK
Post Code	CV47AL
E-mail	[REDACTED]
Telephone	[REDACTED]
Fax	[REDACTED]
Academic supervisor 2	
	Title Forename/Initials Surname Dr Vaso Totsika
Address	CEDAR (Centre for Educational Development Appraisal and Research) University of Warwick Coventry UK
Post Code	CV4 7AL
E-mail	[REDACTED]
Telephone	[REDACTED]
Fax	[REDACTED]

Please state which academic supervisor(s) has responsibility for which student(s):
Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
Student 1 Miss Emma Langley	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika
Student 2 Miss Jane Margetson	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika
Student 3 Miss Mikeda Jess	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- Student
 Academic supervisor
 Other

A3-1. Chief Investigator:

	Title	Forename/Initials	Surname
	Professor	Richard	Hastings
Post	Cerebra Chair of Family Research		
Qualifications	BSc, PhD, CPsychol		

Date: 14/07/2015

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NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

Employer	University of Warwick
Work Address	CEDAR (Centre for Educational Development Appraisal and Research) University of Warwick Coventry UK
Post Code	CV47AL
Work E-mail	[REDACTED]
* Personal E-mail	[REDACTED]
Work Telephone	[REDACTED]
* Personal Telephone/Mobile	[REDACTED]
Fax	[REDACTED]

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.
A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.*

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?
This contact will receive copies of all correspondence from REC and R&D reviewers that is sent to the CI.

	Title	Forename/Initials	Surname
	Mrs	Jane	Prewett
Address	Research Impact Services Coventry		
Post Code	CV4 8UW		
E-mail	WMSponsorship@warwick.ac.uk		
Telephone	[REDACTED]		
Fax	[REDACTED]		

A5-1. Research reference numbers. Please give any relevant references for your study:

Applicant's/organisation's own reference number, e.g. R & D (if available):	R.CDAA.3049
Sponsor's/protocol number:	
Protocol Version:	1.0
Protocol Date:	12/06/2015
Funder's reference number:	N/A
Project website:	

Additional reference number(s):

Ref.Number	Description	Reference Number
	Sponsor reference number	REGO-2015-1604

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

Yes No

Please give brief details and reference numbers.

Date: 14/07/2015

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2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. *Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, this summary will be published on the website of the National Research Ethics Service following the ethical review.*

What are the experiences of UK families with a child with an intellectual disability?

This large-scale longitudinal study investigates the impact on families when they have a child with an intellectual disability. The research is funded by the research charity Cerebra and will recruit 1000 or more families in the UK with a child with an intellectual disability aged 4-11 years. Families will be recruited in a range of ways including social media, communication with special schools, and support from third sector organisations. Families will be asked to participate in Wave 1 of the study (2015-2017) and indicate whether they would also like to be contacted in two years time for Wave 2 (2017-2019).

Wave 1 of the study asks primary and secondary caregivers within each family to report on many aspects of their lives including their own health, well-being and life satisfaction; their children's behaviour and relationships, and general family functioning. The majority of this information will be collected by a self-report online survey, with additional information about the child with the intellectual disability's behaviour collected via a telephone interview (with the primary caregiver).

There is no existing study of this type and size in the UK and so this research will make a significant leap forward in intellectual disability family research and has the potential to lead to decades of high quality family research. Crucially, the outcomes of this research will be important for supporting families. Input from the research charity Cerebra, means that the findings will be made accessible for families and practitioners and will be regularly disseminated at different time points throughout the study.

A6-2. Summary of main issues. *Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.*

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

The physical and ethical risks to the researcher and the participant in this project are minimal because of the use of an online survey method. However, the team is aware that there is a small chance that the survey could cause some emotional distress for caregivers, so have made sure that this is documented in any information prior to consent. As the researcher will not be present when the survey is being completed, it has been ensured that participants know who they can contact if they experience any discomfort or have any concerns. It will be made very clear that they can decide to not complete the survey, or withdraw themselves and any data that they have provided from the study without any reason.

We are also aware that completing the survey will be an additional burden for families. We have tried to negate this in three ways: firstly, by making the survey as short as possible, using essential measures (while still trying to retain the items that will answer the research questions). Secondly, we decided to split the surveys into one online survey and one telephone interview (the interview will only need to be completed by one caregiver if they are willing to do so). Thirdly, participants can fill in the online survey at their own leisure and can opt out of the telephone interview or arrange to complete it at a time that is suitable for them.

A challenge for the project will be to recruit approximately 1000 families, thus the research team has decided to recruit via a number of streams including information on a project website, social media and contact with schools and third sector organisations. Participants will be able to opt-in to the study by clicking through to the survey via the link provided on the study advertisement. If they then decide then that they do not want to proceed, they are under no obligation to continue.

Due to the large-scale nature of the study it is not feasible to adapt the survey or make it available in a language other

than English because of time and cost considerations. While making it available in other languages and mediums is desirable, it would not be possible to adapt the survey for each individual. Additionally, translating existing standardised assessment measures may be difficult due to copyright considerations.

There should be no significant issues for obtaining informed consent because the providing consent is a condition for taking part in the study. Nevertheless we will be providing potential participants with clear yet detailed information on the participation page online. We will make it clear that participants can opt into all stages of the research, including being contacted in two years' time to participate in wave 2 of the project.

Keeping the personal data of participants secure is a key consideration of this study, as the study will be obtaining a wealth of personal information from families including contact and demographic information. This information will only be available to the research team. The electronic data gathered on participants, including personal/demographic information, will be stored securely on firewalled university computers.

The research team will consider the confidentiality of participant data by separating respondents' contact information from their survey responses. These two types of information will be stored on separate secure databases. No individual reporting of results will be provided and practically would not be viable because of the size of the study. Confidentiality will only be broken if a respondent reports something which suggests that they or someone they care for is at risk, and therefore appropriate authorities would be notified. This information will be made clear in the consent procedure.

Any dissemination of results through reports journal articles, conferences or PhD theses will be based on the population of respondents as a whole.

Participants will be made aware that they can view updates and reports from the study via the project website and Facebook and Twitter pages.

A6-3. Proportionate review of REC application *The initial project filter has identified that your study may be suitable for proportionate review by a REC sub-committee. Please consult the current guidance notes from NRES and indicate whether you wish to apply through the proportionate review service or, taking into account your answer to A6-2, you consider there are ethical issues that require consideration at a full REC meeting.*

Yes - proportionate review No - review by full REC meeting

Further comments (optional):

This study raises no significant medical issues, and we believe it is suitable for proportionate review. However, we are building a sample that is likely to be invited to participate in research several times into the future (after the currently proposed Wave 2). Therefore, we are suggesting that a full review would be appropriate.

Note: This question only applies to the REC application.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. Please tick all that apply:

- Case series/ case note review
- Case control
- Cohort observation
- Controlled trial without randomisation
- Cross-sectional study
- Database analysis
- Epidemiology
- Feasibility/ pilot study
- Laboratory study
- Metanalysis
- Qualitative research
- Questionnaire, interview or observation study

NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

- Randomised controlled trial
 Other (please specify)

A10. What is the principal research question/objective? *Please put this in language comprehensible to a lay person.*

We are not distinguishing between one main objective and secondary objectives.

The main aims of this research study are as follows:

1. To describe the well-being of family members of children with intellectual disability and to compare these data wherever possible with normative data and/or to national datasets (especially the Millennium Cohort Study). Family members include mothers (or primary parental caregivers), fathers (or secondary parental caregivers), and siblings.
2. To examine well-being differences between mothers and fathers in families of children with intellectual disability.
3. To explore correlates of maternal, paternal and sibling well-being, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, and the child with intellectual disability's behaviour problems and adaptive skills.
4. To explore correlates of the behavioural and emotional well-being of the child with intellectual disability, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, the child with intellectual disability's adaptive skills, and parenting attitudes and behaviours.

Using a repeated data collection, two years after the initial survey, we will also examine aims 3 and 4 with a focus on longitudinal associations.

A11. What are the secondary research questions/objectives if applicable? *Please put this in language comprehensible to a lay person.*

We are not distinguishing between one main objective and secondary objectives.

The main aims of this research study are as follows:

1. To describe the well-being of family members of children with intellectual disability and to compare these data wherever possible with normative data and/or to national datasets (especially the Millennium Cohort Study). Family members include mothers (or primary parental caregivers), fathers (or secondary parental caregivers), and siblings.
2. To examine well-being differences between mothers and fathers in families of children with intellectual disability.
3. To explore correlates of maternal, paternal and sibling well-being, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, and the child with intellectual disability's behaviour problems and adaptive skills.
4. To explore correlates of the behavioural and emotional well-being of the child with intellectual disability, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, the child with intellectual disability's adaptive skills, and parenting attitudes and behaviours.

Using a repeated data collection, two years after the initial survey, we will also examine aims 3 and 4 with a focus on longitudinal associations.

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

As there has not been a study of this kind on such a large scale in the UK, the findings will be an original contribution to scientific knowledge. To date, intellectual disability (ID) and family research has primarily explored the negative impact of raising a child with ID; however the current study aims to take the position from a positive standpoint, whilst also allowing family members to include any potential negative aspects involved with raising their child. Furthermore, there is limited research on the experiences of the secondary caregivers of children with ID.

This study aims to fill that gap in the scientific literature. The potential findings have both theoretical and practical implications. This large-scale longitudinal study will have a significant impact on intellectual disability research in the UK, with the potential to lead to decades of high quality family research. In addition the findings could identify the key needs of families of children with ID which may lead to the development of practical support and interventions for these families. Thus this research has direct implications for both research and families with an ID child.

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

Date: 14/07/2015

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The large-scale nature of this study necessitates a quantitative design. A qualitative study with one or two home visits to 1,000 families would be prohibitively expensive in human resource and travel costs. The use of a quantitative design is building on previous research in this area which has been small-scale or qualitative in design.

The decision to use an online survey as the main method of data collection reflects the aim to collect vast amounts of data: it will allow for the data to be collected easily at fairly low cost and will reduce the time it takes to complete the research for participants and researchers. The survey also has other benefits in that the measures used can be completed swiftly and can be quantified with ease. Given that we are asking for the participation of families of children with a disability, we feel that the survey will not inconvenience them too much and can be completed in their own homes at their own leisure. The decision to have an online survey and then a telephone interview for the primary caregiver was made to prevent survey 1 becoming too long for the participant to complete. The measures used in the telephone interview can be rapidly completed.

Timetable for participants (Wave 1 only)

1. Participants will see/receive information about the study and can go to the online survey information via the link provided on the project advertisement.
 2. Participants will be asked to indicate on the online survey page whether they are a primary or a secondary caregiver (some definition/guidance is provided) and this will direct them to the full study information and the consent page.
 3. Participants will be required to answer all the consent questions, including whether they wish to be contacted for a telephone interview (Primary caregiver only) and for Wave 2 of the study in two years' time.
 4. Once consent has been completed participants can proceed with the survey.
 5. When a survey has been completed, a researcher will export the information from SurveyMonkey and will extract contact information and arrange a telephone interview with participants (Primary Caregivers only) who stated that they were happy to be contacted.
 6. The telephone interview is then completed (Primary Caregiver only).
 7. Participants who indicate that they would be willing to be invited into Wave 2 of the study will be contacted after approximately two years (by email, telephone or post).
 8. If participants are happy to participate in Wave 2, the process will start again with the consent questions in the online survey.
- If participants have indicated that would like to be kept informed about the results of this study and other family research carried out by the research group, they will be sent a regular e-newsletter. The same information will be available via social media if participants opt not to receive the newsletter.

As the research team will not be meeting any of the participants responding to the survey the influence of researcher bias will be minimal. Nevertheless, in designing the survey the research team have worked together to ensure that the survey is balanced in what it explores and that the measures used have been appraised prior to being included. The use of measures within the study also help to negate any researcher influence, in that they are closed questions and are reported either directly by the respondent online and for the telephone interview marked down on a standardised measure form. In the analysis stage, the research team will work together to analyse the data and PhD students will be guided by their supervisors who will check claims made in their reports and theses.

The main focus of the research is on a large sample to enable more complex research questions to be addressed, and on collecting data about multiple family members. An online survey is the most efficient and effective way to achieve these broader aims. We recognise that the sample obtained will not be representative of the UK population of parents of children with intellectual disability. Other research being carried out by our team addresses this issue of representativeness through secondary analysis of UK population-based datasets. A limitation of existing population-based surveys is the small number of families included who have children with intellectual disability. Thus, our research programme as a whole does address representativeness but this is not a priority for the 1,000 Families Study.

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- Design of the research

- Management of the research
 Undertaking the research
 Analysis of results
 Dissemination of findings
 None of the above

Give details of involvement, or if none please justify the absence of involvement.

We have secured initial Impact funding separate from the research grant to support the recruitment of up to 6 family members to become part of a parent advisory group for this project and to establish themselves as themselves as "Ambassadors" for the research. This role will involve sharing the results with other families, and we will be providing training/mentoring for this group of family members.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

Families with one or more children aged between 4 and 11 years old who have an intellectual disability as reported by a parental caregiver.

Families with at least one parental caregiver responding to the on-line survey.

Families that live in the UK.

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

Families where the child with an intellectual disability lives outside of the family home on a full time basis (e.g. a 52 week residential school placement).

Parental caregivers whose English literacy skills mean they are unable to participate in an on-line and telephone interview.

Parental caregivers under 18 years of age.

RESEARCH PROCEDURES, RISKS AND BENEFITS

A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.

Please complete the columns for each intervention/procedure as follows:

- Total number of interventions/procedures to be received by each participant as part of the research protocol.
- If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
- Average time taken per intervention/procedure (minutes, hours or days)
- Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Consent form	1	0	5 minutes	The participants will complete the consent form as the first section of the online survey. The participant will need to consent before they can continue.
Online Survey	13	0	30-40 minutes	Participants will complete the survey online. 13 different sections are included - demographic questions, and then research measures as specified in the protocol.

NHS REC Form

Reference:
15/WM/0267

IRAS Version 4.0.0

Telephone interview	2 0	35-75 minutes	Education and Psychology PhD students based at the University of Warwick will conduct the telephone interviews from their private office. Participants will be instructed to be in a private place (e.g. a room away from the family/colleagues/other distractions) when the telephone interview takes place. Only the primary caregiver will complete the telephone interview.
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A21. How long do you expect each participant to be in the study in total?

4 years

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

The physical risks to participants are minimal as the research questions are answered via an online survey and telephone interview.

The survey and interview has a very small risk of leading to distress in the participant. Participants will be informed about the nature of the survey/interview before they take part so they are prepared for any feelings that might arise. Before participating, individuals will also be informed of the procedures for contacting the lead researcher/sponsor within a reasonable time, if following participation they experience stress, harm or have any concerns.

The time it will take for individuals to participate in this study may be an additional burden/inconvenience to families of children with an intellectual disability, on top of their caring/other responsibilities. Due care has been given to keep the number of research questions to a minimum and for participants to complete the survey and interview at different times (that suit them), to ensure participation is not overly burdensome.

To minimise these risks we will:

- i) Ensure informed consent
- ii) Ensure PhD students managing the study are supervised and supported appropriately.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

Yes No

A24. What is the potential for benefit to research participants?

The only benefits for participants are the opportunity to contribute to increased understanding of families of children with intellectual disability, and thus to ensure more appropriately designed support services and interventions, and to receive accessible information about the study findings and similar research from our research group and others.

A26. What are the potential risks for the researchers themselves? (if any)

There are no risks to researchers themselves as the survey is conducted online and the interviews are by telephone.

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).

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Participants will be recruited by PhD researchers in the Family Research team. Time and resourcing as part of their PhD is available for them to fulfil this task.

Information about the study will be disseminated through social media activities, such as a project website, Facebook page, and Twitter feed.

Special schools will be contacted and asked to forward information about the study to caregivers.

Recruitment will also be supported by third sector organisations, distributing information through their social media presence and regular newsletters.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

Yes No

Please give details below:

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

Yes No

If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).

A short advert describing the project will be published on-line through a project website and dedicated project Facebook and Twitter site. The same short text will be used by other organisations bringing the study to the attention of families.

A29. How and by whom will potential participants first be approached?

No individual participants will be directly approached by a member of the research team to take part in the study. Individuals who have seen information about the study online or have been directed to the online advert by special schools or third sector organisations will be able to go online to view the full information about the study and participate in the study if they so choose.

A30-1. Will you obtain informed consent from or on behalf of research participants?

Yes No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

The advertisement for the study will provide a link to the first page of the online survey where full information about involvement will be available. If caregivers are happy to participate they can proceed to the consent page where they will be required to read and agree to a series of consent statements before proceeding.

Caregivers will not be able to start the survey if they have not answered the consent questions.

If caregivers have any questions about the project, they will be advised to contact the research team via the project email address provided on the information page.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

Yes No

If No, how will it be recorded?

There will be an electronic record created as a part of the response to the online survey rather than a physical signed consent form.

A31. How long will you allow potential participants to decide whether or not to take part?

There will be no specific time frame imposed on participants to decide whether they take part in the study. They can read the online information about the study at their own leisure and without any commitment to participate.

We anticipate that the survey will be live for between 12 and 18 months.

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?(e.g. translation, use of interpreters)

Caregivers who cannot complete the study because of their English literacy skills or special communication needs will not be involved in the study.

A33-2. What arrangements will you make to comply with the principles of the Welsh Language Act in the provision of information to participants in Wales?

The online and telephone survey will be provided in English only.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.

- The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.
- The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.
- The participant would continue to be included in the study.
- Not applicable – informed consent will not be sought from any participants in this research.
- Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

Due to the large-scale nature of the study, and the non-clinical nature of the sample, the research team will not be able to monitor capacity and therefore will assume continued capacity unless notified otherwise.

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study**A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)?(Tick as appropriate)**

- Access to medical records by those outside the direct healthcare team

- Electronic transfer by magnetic or optical media, email or computer networks
- Sharing of personal data with other organisations
- Export of personal data outside the EEA
- Use of personal addresses, postcodes, faxes, emails or telephone numbers
- Publication of direct quotations from respondents
- Publication of data that might allow identification of individuals
- Use of audio/visual recording devices
- Storage of personal data on any of the following:
 - Manual files including X-rays
 - NHS computers
 - Home or other personal computers
 - University computers
 - Private company computers
 - Laptop computers

Further details:

Researchers will use email as one of the media to communicate with participants, using email addresses that the parents have provided - to answer questions about the study, to make arrangements for telephone interviews, and to disseminate study results to parents. Emails with parents will not be used as a means to send or receive personal data.

Participants personal information including names and contact information is required to contact participants for the follow up study if they have agreed to this on the consent form, and to ensure that we can match together parents from the same family (required for analysis).

If participants would like to be kept up-to-date with the research findings we will also use their contact details to do this. Participants' telephone numbers will also be used if they have agreed to participate in the telephone interview stage of the study.

Personal data will be stored electronically on fire-walled University computers. These computers will be password protected and only accessible by the named researchers and the Chief Investigator.

Anonymised data only will be downloaded to personal computers or laptops for the purposes of data analysis. Data for analysis will include identifying participant numbers but the database linking these numbers to personal information will not be downloaded or transferred to personal or laptop computers. All personal and laptop computers will be password-protected as will the folders containing any data analysis file.

A38. How will you ensure the confidentiality of personal data? *Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.*

Participants will be assigned a randomised number to maintain anonymity.

Participants' responses will be pooled together with other participants for analyses so that that individual responses cannot be identified in any way.

Participants' names and personal information will not be able to be identified in any report or presentation about the study.

A40. Who will have access to participants' personal data during the study? *Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.*

The named PhD researchers, (Mikeda Jess, Emma Langley and Jane Margetson) and their supervisors (Prof Richard Hastings and Dr Vaso Totsika), and part-time research assistant Liz Halstead.

Storage and use of data after the end of the study

A43. How long will personal data be stored or accessed after the study has ended?

- Less than 3 months
 3 – 6 months
 6 – 12 months
 12 months – 3 years
 Over 3 years

If longer than 12 months, please justify:

Data collected for this study is to be used as part of an on going longitudinal study. Permission from participants will be granted in the consent procedure to be invited to participate in a follow-up survey 2 years from the initial survey.

INCENTIVES AND PAYMENTS**A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?**

- Yes No

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

- Yes No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

- Yes No

NOTIFICATION OF OTHER PROFESSIONALS**A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?**

- Yes No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION**A50. Will the research be registered on a public database?**

- Yes No

Please give details, or justify if not registering the research.

This study does not involve the trial of an intervention. Cerebra, as the funding organisation, will include information about the study and its progress on their website.

Registration of research studies is encouraged wherever possible.

You may be able to register your study through your NHS organisation or a register run by a medical research charity,

or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A51. How do you intend to report and disseminate the results of the study? Tick as appropriate:

- Peer reviewed scientific journals
- Internal report
- Conference presentation
- Publication on website
- Other publication
- Submission to regulatory authorities
- Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- No plans to report or disseminate the results
- Other (please specify)

A53. Will you inform participants of the results?

Yes No

Please give details of how you will inform participants or justify if not doing so.

Participants will not be informed about their individual survey results as they will be pooled together with those of other participants for analysis.

The research team will be making regular updates on the project website and Facebook and Twitter pages. Participants can be kept up-to-date with the research by accessing these pages. For participants who have indicated their willingness to receive regular electronic updates, we will send them regular e-newsletters about the results and the findings from similar research.

5. Scientific and Statistical Review

A54. How has the scientific quality of the research been assessed? Tick as appropriate:

- Independent external review
- Review within a company
- Review within a multi-centre research group
- Review within the Chief Investigator's institution or host organisation
- Review within the research team
- Review by educational supervisor
- Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

The proposal for this research study was reviewed initially by the internal research team and Trustees of the funding charity Cerebra as a part of the process to decide whether to award funding to Professor Hastings as a Cerebra Academic Chair. The detailed protocol was reviewed and developed by Professor Hastings and Dr Vaso Totsika as supervisors of three PhD students who will be responsible for working together to recruit to this study and to analyse key data.

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

A56. How have the statistical aspects of the research been reviewed? *Tick as appropriate:*

- Review by independent statistician commissioned by funder or sponsor
 Other review by independent statistician
 Review by company statistician
 Review by a statistician within the Chief Investigator's institution
 Review by a statistician within the research team or multi-centre group
 Review by educational supervisor
 Other review by individual with relevant statistical expertise
 No review necessary as only frequencies and associations will be assessed – details of statistical input not required

In all cases please give details below of the individual responsible for reviewing the statistical aspects. If advice has been provided in confidence, give details of the department and institution concerned.

	Title	Forename/Initials	Surname
	Dr	Vaso	Totsika
Department	Centre for Educational Development, Appraisal and Research (CEDAR)		
Institution	University of Warwick		
Work Address	Coventry		
Post Code	CV47AL		
Telephone	024 765 22185		
Fax			
Mobile			
E-mail			

Please enclose a copy of any available comments or reports from a statistician.

A57. What is the primary outcome measure for the study?

The primary outcome for this study depends on which of several different research aims is being addressed. For some analyses, the outcomes of interest will be parental well-being (e.g., stress, mental health, positive perceptions), and for others the outcomes will be child behavioural and emotional problems or adaptive skills.

A58. What are the secondary outcome measures? *(if any)***A59. What is the sample size for the research?** *How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.*

Total UK sample size:	4000
Total international sample size (including UK):	4000
Total in European Economic Area:	0

Further details:

Our aim is to recruit at least 1000 (and up to 2000) families of children with an intellectual disability. If two parental caregivers responded from 2000 families, the maximum number of participants would be 4000.

A60. How was the sample size decided upon? *If a formal sample size calculation was used, indicate how this was done,*

giving sufficient information to justify and reproduce the calculation.

The target number of families is set to at least 1000, in part because the aim is for this to be the largest survey on families of a child with an intellectual disability in the UK. Importantly, a large sample size in Wave 1 is imperative to ensure a good sample size in Wave 2, to allow for attrition.

To examine the power of the proposed (minimum) sample size of 1,000 families, we need to consider (a) the nature of the data, and (b) the most analytically complex and demanding models that will be employed to address the research aims.

All data collected will come from families and will refer to at least two family members (e.g., mothers-fathers, mother-child; mother-sibling). Any type of modelling will need to account for the clustering of participants within families. At the micro-level (Level 1) we have data for each individual family member (child, mother, father, sibling). At the macro-level (Level 2) we have information about the family as a whole (e.g., family income). Individuals who cluster within the same level (i.e., family) are expected to have outcomes more similar than individuals from different clusters. We, therefore, need to account for this potential association in all our models.

There are two multivariate approaches that are appropriate for this type of data: (a) multi-level models (also known as mixed models), and (b) generalised estimating equations. Both will be used to address research questions in this project. They are appropriate for analysing clustered data, and are a type of hierarchical regression analysis. These models will explore associations between child, mother, father, sibling, and overall family environment factors (e.g., family economic position) associated with child outcomes (e.g., child behaviour problems). Standard regression models cannot be fitted to this type of data as the hierarchical nature of the data violates the assumption of independence (as noted above), whereas multi-level models not only accommodate this but treat it as a substantive part of the analysis. Generalised estimating equations can accommodate the issue of clustering by accounting for its effect on the substantive part of the modelling. Power analyses for generalised estimating equations in family research have not been well developed yet, but the rationale and guidance will be similar to that for multi-level models, with one caveat. Available information on linear multilevel models indicates that sample size is equally important at every level of the investigation, and for this we will aim to achieve a large sample of participating families and children (i.e., sample sufficient numbers at the micro and macro level). Available guidance for generalised estimating equations (Garson, 2013) indicates that sample size is more important for level 2 (i.e., the macro-level) variables. For the present design (one child with intellectual disability recruited per family), this suggests that Level 2 and Level 1 sample sizes should be targeted at the same rate (i.e., if we need 1,000 children we need to target 1,000 families, and vice versa).

Available guidance on linear 2-level multilevel models suggests that power of about .80 for level 1 fixed effect predictors can be achieved by samples of between 600 to 1200 level-1 participants (Bell et al., 2010). Assuming more demanding models with 20 covariates, and low levels of intra-class correlation (0.10; a reasonable expectation on the basis of our previous research; Jones et al., 2013), 1,000 level 1 participants would allow power of about 1.0 to detect a small effect size (.30) in a 2-level design (Hedges & Rhoads, 2010). Therefore, the power analysis suggests that a proposed sample size of 1,000 level 1 participants is powerful enough to detect at least a small effect size in presence of several model covariates and low intra-class correlation. In this way, power for analytically less demanding models will also be sufficient.

A61. Will participants be allocated to groups at random?

Yes No

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

SPSS software will be used to perform statistical analysis according to the research aims.

Levels of well-being in mothers, fathers and siblings will be analysed through descriptive statistics and one-sample t-tests (and non-parametric alternatives where appropriate). This will be compared to levels of well-being in families from normative data and other UK samples.

Multi-level modelling will be used to explore well-being scores, relationship quality, and parenting attitudes and beliefs between mothers and fathers in the same families.

Factors associated with the well-being of mothers, fathers and siblings will be explored through correlation and regression approaches. This will also be used to explore the factors associated with the well-being of the child with the intellectual disability. Final models will be tested using structural equation modelling.

Structural equation modelling approaches will be used for the longitudinal analyses to focus on factors associated with the well-being of the child with the intellectual disability and family members over time.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.

	Title	Forename/Initials	Surname
	Ms	Elizabeth	Halstead
Post	Visiting Fellow		
Qualifications	BSc Psychology		
Employer	Cerebra		
Work Address	CEDAR		
	University of Warwick		
Post Code	CV4 7AL		
Telephone			
Fax			
Mobile			
Work Email	[REDACTED]		

	Title	Forename/Initials	Surname
Post			
Qualifications			
Employer			
Work Address			
Post Code			
Telephone			
Fax			
Mobile			
Work Email			

	Title	Forename/Initials	Surname
Post			
Qualifications			
Employer			
Work Address			
Post Code			
Telephone			
Fax			
Mobile			

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Work Email
Title Forename/Initials Surname
Post
Qualifications
Employer
Work Address
Post Code
Telephone
Fax
Mobile
Work Email

A64. Details of research sponsor(s)

A64-1. Sponsor

Lead Sponsor

- Status: NHS or HSC care organisation
 Academic
 Pharmaceutical industry
 Medical device industry
 Local Authority
 Other social care provider (including voluntary sector or private organisation)
 Other
- Commercial status: Non-Commercial

If Other, please specify:

Contact person

Name of organisation University of Warwick
 Given name Jane
 Family name Prewett
 Address Research and Impact Services
 Town/city Coventry
 Post code CV4 8UW
 Country UNITED KINGDOM
 Telephone [REDACTED]
 Fax
 E-mail WMSSponsorship@warwick.ac.uk

Is the sponsor based outside the UK?

- Yes No

Under the Research Governance Framework for Health and Social Care, a sponsor outside the UK must appoint a

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legal representative established in the UK. Please consult the guidance notes.

A65. Has external funding for the research been secured?

- Funding secured from one or more funders
 External funding application to one or more funders in progress
 No application for external funding will be made

What type of research project is this?

- Standalone project
 Project that is part of a programme grant
 Project that is part of a Centre grant
 Project that is part of a fellowship/ personal award/ research training award
 Other

Other – please state:

Please give details of funding applications.

Organisation Cerebra
Address 2nd Floor Offices, The Lyric Buildings
King Street
Carmarthen
Post Code SA31 1BD
Telephone 01267244200
Fax
Mobile
Email

Funding Application Status: Secured In progress

Amount: 270,000

Duration

Years: 6

Months:

If applicable, please specify the programme/ funding stream:

What is the funding stream/ programme for this research project?

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

- Yes No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

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A69-1. How long do you expect the study to last in the UK?

Planned start date: 01/09/2015

Planned end date: 31/12/2019

Total duration:

Years: 4 Months: 3 Days: 31

A71-2. Where will the research take place? (Tick as appropriate)

- England
 Scotland
 Wales
 Northern Ireland
 Other countries in European Economic Area

Total UK sites in study 1

Does this trial involve countries outside the EU?
 Yes No
A72. What host organisations (NHS or other) in the UK will be responsible for the research sites? Please indicate the type of organisation by ticking the box and give approximate numbers of planned research sites:

- NHS organisations in England
 NHS organisations in Wales
 NHS organisations in Scotland
 HSC organisations in Northern Ireland
 GP practices in England
 GP practices in Wales
 GP practices in Scotland
 GP practices in Northern Ireland
 Social care organisations
 Phase 1 trial units
 Prison establishments
 Probation areas
 Independent hospitals
 Educational establishments 1
 Independent research units
 Other (give details)

Total UK sites in study: 1

A76. Insurance/ indemnity to meet potential legal liabilities

Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.

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Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (NHS sponsors only)
 Other insurance or indemnity arrangements will apply (give details below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (protocol authors with NHS contracts only)
 Other insurance or indemnity arrangements will apply (give details below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

- NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
 Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

Please enclose a copy of relevant documents.

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

Research site		Investigator/ Collaborator/ Contact	
Institution name	University of Warwick	Title	Professor
Department name	Centre for Educational Development Appraisal and Research	First name/ Initials	Richard
Street address	University of Warwick	Surname	Hastings
Town/city	Coventry		
Post Code	CV4 7AL		

PART D: Declarations**D1. Declaration by Chief Investigator**

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
3. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
4. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
5. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
6. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
7. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
8. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 1998.
9. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
 - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
 - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
 - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
 - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
 - May be sent by email to REC members.
10. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
11. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Contact point for publication*(Not applicable for R&D Forms)*

NRES would like to include a contact point with the published summary of the study for those wishing to seek further information. We would be grateful if you would indicate one of the contact points below.

- Chief Investigator
 Sponsor

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- Study co-ordinator
- Student
- Other – please give details
- None

Access to application for training purposes *(Not applicable for R&D Forms)**Optional – please tick as appropriate:*

I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

This section was signed electronically by Professor Richard Hastings on 13/07/2015 15:03.

Job Title/Post: Professor
Organisation: University of Warwick
Email: [REDACTED]

DRAFT

D2. Declaration by the sponsor's representative

If there is more than one sponsor, this declaration should be signed on behalf of the co-sponsors by a representative of the lead sponsor named at A64-1.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.

Please note: The declarations below do not form part of the application for approval above. They will not be considered by the Research Ethics Committee.

6. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.
7. Specifically, for submissions to the Research Ethics Committees (RECs) I declare that any and all clinical trials approved by the HRA since 30th September 2013 (as defined on IRAS categories as clinical trials of medicines, devices, combination of medicines and devices or other clinical trials) have been registered on a publically accessible register in compliance with the HRA registration requirements for the UK, or that any deferral granted by the HRA still applies.

This section was signed electronically by Mrs Jane Prewett on 13/07/2015 23:33.

Job Title/Post: Deputy Director, R&IS

Organisation: University of Warwick

Email: [REDACTED]

D3. Declaration for student projects by academic supervisor(s)

1. I have read and approved both the research proposal and this application. I am satisfied that the scientific content of the research is satisfactory for an educational qualification at this level.
2. I undertake to fulfil the responsibilities of the supervisor for this study as set out in the Research Governance Framework for Health and Social Care.
3. I take responsibility for ensuring that this study is conducted in accordance with the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research, in conjunction with clinical supervisors as appropriate.
4. I take responsibility for ensuring that the applicant is up to date and complies with the requirements of the law and relevant guidelines relating to security and confidentiality of patient and other personal data, in conjunction with clinical supervisors as appropriate.

Academic supervisor 1

This section was signed electronically by Dr Vasiliki Totsika on 13/07/2015 21:33.

Job Title/Post: Senior Research Fellow

Organisation: University of Warwick

Email: [REDACTED]

Academic supervisor 2

This section was signed electronically by Professor Richard Hastings on 13/07/2015 15:04.

Job Title/Post: Professor

Organisation: University of Warwick

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DRAFT

4.2: Research Sponsor Approval Letter

Warwick Medical School

7th July 2015

Prof Richard Hastings
CEDAR
University of Warwick
Coventry
CV4 7AL
United Kingdom

Project Title: The 1000 Families Study
Chief Investigator: Richard Hastings
Our Ref: REGO-2015-1604

Dear Professor Hastings,

I confirm that the University of Warwick will act as Research Sponsor for the above project, in accordance with the Department of Health's Research Governance Framework for Health and Social care (2005), and, where appropriate, UK Statutory Instrument Number 1031, that implements the Medicines for Human Use (Clinical Trials) Directive 2004 and subsequent amendments.

I confirm that the University holds Public and Products Liability Insurance, and, where appropriate, Clinical Trial Insurance, which will provide cover for this study.

Any researcher involved in the project is required at all times to comply with the University of Warwick's Research Code of Practice.

Best wishes



Graham Hewitt
Research Ethics and Governance Manager

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4.3: 1000 Families Protocol – Final Version

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The 1,000 Families study: Well-being in families of children with intellectual disability

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Funder and reference number:	Cerebra, and ESRC Doctoral Training Centre, University of Warwick
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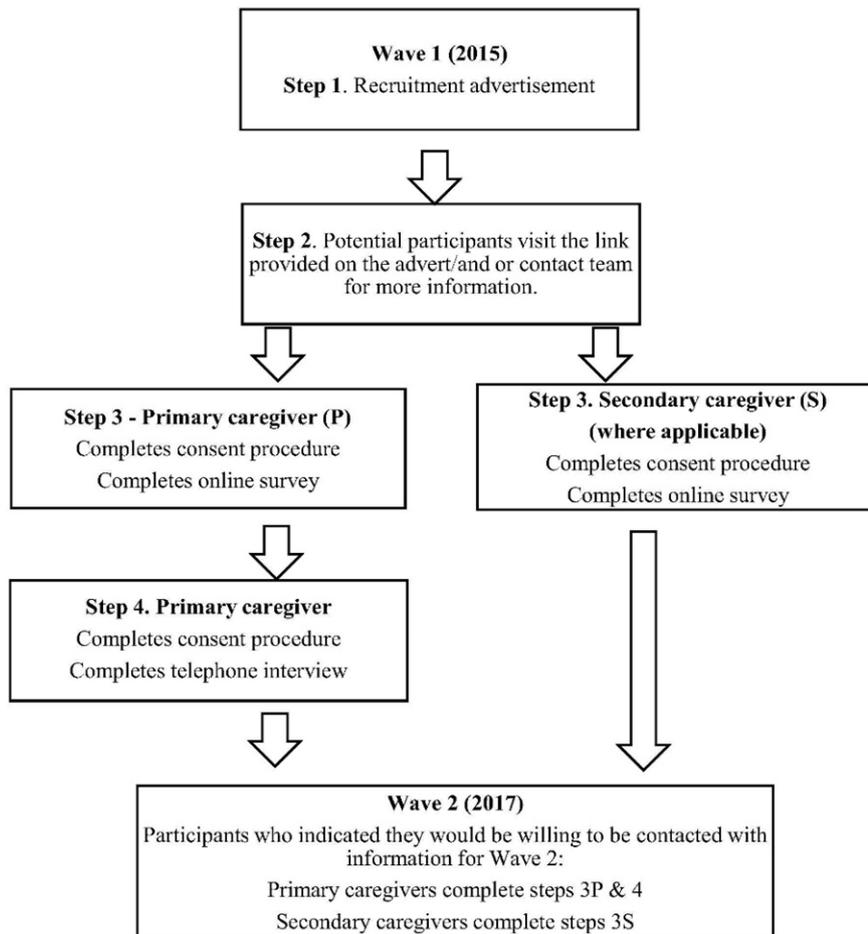
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1. Outline of Study - flow chart and Summary

1.1 Figure 1 study flow chart



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1.2 Summary

“*The 1,000 Families Study*” focuses on families of children and young people with intellectual disability (ID), including autism. There is no existing large-scale (more than 100-200 families) ongoing longitudinal study of the families of children with ID or autism in the UK.

We will recruit a UK sample of at least a 1000 (and up to 2000) families living with a child with intellectual disability (including autism) of school age (11 years of age in 2015/16). Data will be collected through an online survey and a telephone interview. Initial data collection will take place in 2015-16, with repeat data collection two years later in 2017-19. The measures will focus on the wellbeing of the child, their parents and also their siblings.

2. Background

Parents, especially mothers, raising children with ID or autism report more stress and mental health difficulties than do other parents. Our research group has carried out secondary analysis of large-scale UK databases where the sampling was representative of the UK population of children (i.e., not based on sampling families only via contact with services or from samples of parents volunteering to participate in a study). Our analyses show that mothers’ psychological well-being is poorer when they have a child with ID and/or autism even when you control for other important variables such as socio-economic circumstances (Totsika et al., 2011a).

2.1 The impact of behaviour problems

Many research studies make it clear that one variable that explains why mothers in particular of children with ID and autism experience more stress is their children’s increased behaviour problems compared to other children (Hastings, 2002). In addition, the relationship between child behaviour problems and maternal well-being seems to be bidirectional – maternal stress is driven by children’s behaviour problems, but also children of stressed mothers are likely to have worsening behaviour problems over time (Hastings et al., 2006). There is still a research need to explore these bidirectional relationships in more detail.

2.2 Variation in parental stress

Although there is clear evidence that parents report more stress when they have a child with ID or autism, many parents simply do not report heightened levels of psychological distress. For example,

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in our recent secondary analysis research approximately 40% of mothers reported emotional problems at a concerning level and 60% did not (Totsika et al., 2011a). An important question in terms of family strengths is why some parents can cope with significant challenges when others seem less able to do so at a particular point in time. One factor may relate a family's life stage: periods around initial diagnosis and transition to adulthood may be especially challenging for parents. A second issue is that family economic resources are clearly influential, explaining a significant proportion of the variation in maternal psychological well-being (Totsika et al., 2011a). Families living in deprived economic circumstances have additional stresses, have fewer resources, and often have fewer skills to advocate for their child and family.

2.3 Fathers, siblings and extended family

Few studies have investigated the effect of having a child with an intellectual disability on members of the family other than mothers – such as fathers and siblings. While some research has reported that fathers of children with ID and autism also generally report increased stress compared to other fathers (MacDonald & Hastings, 2010), research findings are by no means consistent. Fathers experience raising a child with ID or autism differently than do mothers (Jones et al., 2013) including reporting less stress overall than their partners. Although parents are often worried about the negative impact on siblings of having a brother or sister with ID or autism, research data suggest that the impact on sibling psychological well-being is only very small overall (Petalas et al., 2009a) and can even be positive in the form of, for example, increased sensitivity/tolerance for difference (Petalas et al., 2009b, 2012a).

Consistent with research findings from studies of mothers, fathers and siblings also report more psychological problems when the child with ID or autism has significant behaviour problems (Hastings, 2007; Petalas et al., 2012b).

3. Research aims

The main aims of this research study are as follows:

1. To describe the well-being of family members of children with intellectual disability and to compare these data wherever possible with normative data and/or to national datasets (especially the Millennium Cohort Study). Family members include mothers (or primary parental caregivers), fathers (or secondary parental caregivers), and siblings.
2. To examine well-being differences between mothers and fathers in families of children with intellectual disability.

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3. To explore correlates of maternal, paternal and sibling well-being, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, and the child with intellectual disability's behaviour problems and adaptive skills.
4. To explore correlates of the behavioural and emotional well-being of the child with intellectual disability, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, the child with intellectual disability's adaptive skills, and parenting attitudes and behaviours.

Using a repeated data collection, two years after the initial survey, we will also examine aims 3 and 4 with a focus on longitudinal associations.

4. Study design

To be able to address all four main research aims, we will conduct an online cross-sectional survey and telephone interview study of at least 1,000 families of children with intellectual disability.

To examine longitudinal associations for research aims 3 and 4, we will invite families to participate in further data collection. At this stage, we are seeking ethical approval for the first two waves of data collection only. During the consent procedure, participants will be asked whether they are happy to be contacted for Wave 2 of the data collection.

5. Sample and recruitment

5.1 Recruitment strategy

We aim to recruit at least 1,000 (and up to 2,000) families of children with an intellectual disability (including children with autism who also have intellectual disability). Recruitment will involve a multi-point method. The main method will involve promotion of the study online through the creation of a project website and social networking pages including Facebook and Twitter. An advert will be placed on these sites which will briefly describe what the research is about and will provide a direct web link to the information and consent page of the online survey.

Recruitment will also include mail-outs to organisations and parents of children with intellectual disability via organisations such as the Learning Disability and Autism Research Network West

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Midlands (LeARN-WM)¹ whose members have agreed to receive information about relevant research projects. Assistance in publicising the research will be sought from other third sector and social care organisations to help identify families and send out information via email, newsletters and their own social media activities. Letters and email communication will also be sent to special schools requesting for information about the study to be passed on to parents.

5.2 Target population

Families of children with an intellectual disability are the target participants for this research. Within each family, data will be gathered from at least one parental caregiver and ideally from both a primary and secondary caregiver (i.e., often, mothers and fathers) where available. The families recruited must have a child aged **between 4 and 11 years of age** with an intellectual disability. 

5.3 Inclusion criteria

1. For initial recruitment, families with one, or more children aged **between 4 and 11 years old**  who have an intellectual disability as reported by a parental caregiver
2. Families with at least one parental caregiver responding to the survey
3. Families that live within the UK

5.4 Exclusion criteria

1. For initial recruitment, families in which the child with intellectual disability lives outside of the family home on a full time basis (e.g., in a 52 week residential school placement)
2. Parental caregivers whose English literacy skills mean that they are unable to participate in an online survey and telephone interview
3. Parental caregivers under 18 years old

6. Recruitment process

6.1 Number of participants

¹ <http://www.birmingham.ac.uk/schools/psychology/centres/cerebra/news-events/Learn-WM-News.aspx>

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A minimum of 1,000 and a maximum of 2,000 families will be recruited. The total number of participants will be a maximum of 4,000 based on two parental caregivers participating from every family – which is a very unlikely outcome. Participants are primary and secondary parental caregivers (parents) of children with intellectual disability.

The target number of families is set to at least 1000, in part because the aim is for this to be the largest survey on families of a child with an intellectual disability (including autism) in the UK. Importantly, a large sample size in wave one is imperative to ensure a good sample size in wave two, to allow for attrition.

To examine the power of the proposed (minimum) sample size of 1,000 families, we need to consider (a) the nature of the data, and (b) the most analytically complex and demanding models that will be employed to address the research aims.

All data collected will come from families and will refer to at least two family members (e.g., mothers-fathers, mother-child; mother-sibling). Any type of modelling will need to account for the clustering of participants within families. At the micro-level (Level 1) we have data for each individual family member (child, mother, father, sibling). At the macro-level (Level 2) we have information about the family as a whole (e.g., family income). Individuals who cluster within the same level (i.e., family) are expected to have outcomes more similar than individuals from different clusters. We, therefore, need to account for this potential association in all our models.

There are two multivariate approaches that are appropriate for this type of data: (a) multi-level models (also known as mixed models), and (b) generalised estimating equations. Both will be used to address research questions in this project. They are appropriate for analysing clustered data, and are a type of hierarchical regression analysis. These models will explore associations between child, mother, father, sibling, and overall family environment factors (e.g., family economic position) associated with child outcomes (e.g., child behaviour problems). Standard regression models cannot be fitted to this type of data as the hierarchical nature of the data violates the assumption of independence (as noted above), whereas multi-level models not only accommodate this but treat it as a substantive part of the analysis. Generalised estimating equations can accommodate the issue of clustering by accounting for its effect on the substantive part of the modelling. Power analyses for generalised estimating equations in family research have not been well developed yet, but the rationale and guidance will be similar to that for multi-level models, with one caveat. Available information on linear multilevel models indicates that sample size is equally important at every level of the investigation, and for this we will aim to achieve a large sample of participating families and children (i.e., sample sufficient numbers at the micro and macro level). Available guidance for generalised estimating equations (Garson, 2013) indicates that sample size is more important for level 2 (i.e., the macro-level) variables. For the present design (one child with intellectual disability recruited per family), this suggests that Level 2

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and Level 1 sample sizes should be targeted at the same rate (i.e., if we need 1,000 children we need to target 1,000 families, and vice versa).

Available guidance on linear 2-level multilevel models suggests that power of about .80 for level 1 fixed effect predictors can be achieved by samples of between 600 to 1200 level-1 participants (Bell et al., 2010). Assuming more demanding models with 20 covariates, and low levels of intra-class correlation (0.10; a reasonable expectation on the basis of our previous research; Jones et al., 2013), 1,000 level 1 participants would allow power of about 1.0 to detect a small effect size (.30) in a 2-level design (Hedges & Rhoads, 2010). Therefore, the power analysis suggests that a proposed sample size of 1,000 level 1 participants is powerful enough to detect at least a small effect size in presence of several model covariates and low intra-class correlation. In this way, power for analytically less demanding models will also be sufficient.

6.2 Informed consent

Informed consent will be obtained from each participant, separately for each step of the study. In Wave 1 each participant will be provided with information about the study on the first screen of the online survey. Participants will be asked to indicate their consent online before completing the survey by responding to a series of consent questions. To proceed to the survey, participants must confirm that they have read and understood the information provided about the study, understand that they can withdraw at any time, understand that their data will be treated with confidence, and agree to participate in the online survey. Participants will also be asked to indicate if they want to take part in the telephone survey and if they are willing to be contacted again in approximately two years' time to consider taking part in the second wave of the study. The researchers will note this information on a separate database so that those that indicate that they want to be contacted later are identifiable.

It will also be made clear that contact/demographic information about themselves and the child with the disability will need to be provided in the survey to match caregiver responses from the same family. This will include the caregivers' full name, home and mobile telephone number, an email address and postal address; and their child's full name and date of birth. This information will be kept secure in a separate database from their survey responses.

Participants will be able to contact the research team with any questions or queries before they participate. An email address and telephone number for the team will be provided on the information sheet on the first screen of the survey.

For Wave 2 of the data collection, participants that have indicated that they are willing for the research team to contact them again will be contacted after two years by email, telephone or mail.

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Consent will be taken as it was in Wave 1 (via the online survey) and participants will be asked again if they would be willing to be contacted for any future research. It will be made clear that there will be no obligation to participate in Wave 2 of the data collection even if participants indicate on the consent form in Wave 1 that they were happy to be contacted again.

7. Withdrawal

Participants have the right to withdraw consent for participation in the study at any time. They do not have to provide a reason for withdrawal. Data will be deleted from the study dataset for any parent who withdraws from the study. If a parent decides to withdraw from the study at the second data collection point, a researcher will check whether they wish to withdraw entirely from the study (in which case all data will be deleted) or only from the follow-up data collection (in which case data from the second wave of data collection only will be deleted).

8. Methodology

8.1 Measures/assessment instruments

The primary and secondary (if available) parental caregiver will be asked to complete the same online survey (see Table 1 for domains/variables and measures). The online survey will be hosted on SurveyMonkey. In addition to the measures listed in Table 1, the participants will first complete a section which gathers demographic information about themselves and their children and family. Demographic variables will include:

- Caregiver name and contact details (postal address, telephone number, email address)
- First, last name and date of birth of the child with the intellectual disability
- Primary/secondary carer gender, marital status, ethnic group, level of education, health/disability
- Number of adults and children in household
- Household income and financial hardship
- Information about the child with the intellectual disability (e.g., diagnoses)
- Information about where the child with the intellectual disability lives during a normal week and where they go to school

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Table 1: Online survey measures

Domain/variable	Participant measures
Parental positivity	Positive gains scale Pit-ten Cate (2003) 7 items
Parent mental health	Kessler 6 (K6) Kessler et al (2003) 6 items
Carer burden/stress	'Impact of care-giving on carer' Survey of Informal Carers in Households 2009/2010 7 items
Life satisfaction	Life satisfaction Department for Environment, Food and Rural Affairs (DEFRA) (2011) 1 item
Parent relationship – parent's perception of relationship with their partner (if relevant)	Disagreement over issues related to child Millennium Cohort Study (2003-2005) 1 item Happiness of relationship scale Millennium Cohort Study Wave 2 (2003-2005) 1 item
Overall perception of family functioning	Family APGAR scale Smilkstein (1978) 5 items
Child with intellectual disabilities behavioural and emotional well being.	Strengths and difficulties questionnaire (SDQ) (Parent questionnaire) Goodman (1997) 25 items
Sibling behavioural and emotional well-being – siblings closest in age to the child with intellectual disability	Strengths and difficulties questionnaire (SDQ) (Parent questionnaire) Goodman (1997) 25 items
Sibling relationship	Sibling relationship questionnaire (SRQ) (Parent questionnaire) (revised) Furman and Buhrmester (1985) 10 items
Parenting attitudes and behaviours	Child-parent relationship scale (CPRS) Pianta (1995) 15 items Alabama Parenting questionnaire (Parent form)

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	Frick (1991) 12 items Parent activities/involvement index. 5 items
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The primary parental caregiver will also be invited take part in a telephone survey following the online/postal survey to provide information about the skills and behaviours of their child with ID (Table 2).

Table 2: Telephone survey measures

Domain/Variable	Participant measures
Behavioural and emotional problems of the child with intellectual disability	Development behaviour checklist (parent version) 96 items (Einfeld & Tonge, 2002)
Adaptive behaviour skills of the child with intellectual disability	Vineland adaptive behaviour scales – second edition (parent interview form) 15-20 min interview Sparrow, Balla & Cicchetti (2005)

8.1.1 Parental positivity

The Positive Gains Scale (Pit-ten-Cate, 2003) is a 7 item measure which requires parents of disabled children to rate the positive impact their disabled child has had on themselves and their family. Five items relate to the perceived benefits for the parent of raising a child with a disability (e.g. ‘Since having this child I have a greater understanding of other people’) and 2 on what the family has gained (e.g. ‘Since having this child, my family has become more tolerant and accepting’). Respondents are required to agree or disagree with each of the 7 statements on a 5-point agreement scale from ‘0=strongly agree’ to ‘4=strongly disagree’. The lower the score, the higher the positive gains reported by parents. The Positive Gains Scale has been used successfully by MacDonald, Hastings and Fitzsimons (2009) on fathers of children with an intellectual disability with a Cronbach’s alpha coefficient of 0.80.

8.1.2 Parental mental health

The Kessler (K6) Scale (Kessler et al, 2003) is a 6 item measure that screens for serious mental illness (SMI), i.e., any DSM-IV disorder other than substance abuse. The 6 questions are mandatory and requires the respondent to rate how often they have felt a frequency of symptoms (e.g. nervousness, worthlessness etc.) within the last 30 day on a 5-point Likert scale (All of the time/ some of the time/

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A little of the time/ None of the time). Total scores range from 6 (indicating no distress) to 30 (indicating severe distress). A cut-off of 8-12 has been suggested for screening for mild-moderate mental health problems, and a score of 13 and above is reflective of serious mental illness (Kessler et al., 2003). The K6 has been used successfully by Weiss and Lunsby (2011) to screen for mental health problems in parents of adults and young children (2-46 years of age) with Autism with a Cronbach's alpha coefficient of 0.87.

8.1.3 Carer burden/stress

Impact on care-giving on carer scale (Survey of informal carers in households 2009/10): This is a 7-item scale that asks those that care for an individual to indicate whether certain aspects of their lives have been affected by caring for another. This includes reduced time socialising, with partner, friends, and doing sport or hobbies. Parents will be required to select 'Yes' or 'No' to the items. There are no cut-off scores available for this measure. Totals will be generated for the yes and no responses.

8.1.4 Life satisfaction

Life satisfaction scale (DEFRA, 2011): This is a single item measure that asks parents to rate their general life satisfaction on a scale of 1 (completely dissatisfied) to 10 (completely satisfied). This scale has been used within major UK social surveys with large-normative samples (n=1000+), including the Survey of Public Attitudes and Behaviour towards the environment by the Department for Environment, Food and Rural Affairs (DEFRA) (2011). Scores between 6 and 10 indicate that respondents are 'satisfied overall'. Scores between 0 and 4 indicate that participants are 'dissatisfied overall'.

8.1.5 Parent relationship

Happiness and disagreement: These are two separate items to be self-completed by each parent (where they are in a relationship). The first asks for the respondent to rate how happy they are with their relationship with their partner from 1 (Very unhappy) to 7 (Very happy). The second asks parents to report on how often they disagree over issues relating to their child on a 7-point Likert scale (Never/Less than once a week/Once a week/Several times a week/Once a day/More than once a day/Can't say). There are no cut-off scores for these items. Answers will be totalled.

8.1.6 Family satisfaction

The Family Apgar Scale (Smilkstein, 1978) is a 5 item measure that assesses a family member's perception of family functioning by examining his/ her satisfaction with family relationships. The measure consists of 5 parameters of family functioning: Adaptability, Partnership, Growth, Affection

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and Resolve (APGAR). Respondents are required to rate the frequency of feeling satisfied with each parameter on a 3-point Likert scale, '0=hardly ever', '1= some of the time', '2= almost always.' The scale is scored by summing the values for the 5 items for a total score that can range from 0- 10. A higher score indicates a greater degree of satisfaction with family functioning (8-10=highly functional, 4-7= moderately dysfunctional, 0-3= dysfunctional).

8.1.7 Siblings

Strengths and difficulties questionnaire (SDQ) (parent version) (Goodman, 1997): This is a 25-item behavioural screening questionnaire completed by parental caregivers about a child aged between 4 and 16 years. Goodman and Goodman (2009) have demonstrated that children who have higher difficulty scores have greater psychopathology as judged by the prevalence of clinical disorder. It uses 5 key scales each with 5 items: 1) emotional problems, 2) conduct problems, 3) hyperactivity/inattention, 4) peer relationship problems, and 5) prosocial behaviour. A total difficulties score can be obtained by adding the scores from scales 1-4 (20 items) and categorising them in new four band system: (0-13), 'close to average', (14-16), 'slightly raised' (17-19) 'High' and (20-40). 'Very high'

Sibling relationship questionnaire (SRQ-SF) (short form adapted) (Furman and Buhrmester 1985) This questionnaire in its short-form is 39 items in length with parents reporting on the connections and tensions among siblings in the family. We have selected two key subscales from the SRQ-SF: warmth and closeness (6 items) and conflict (4 items) to create a 10 item measure. Parents are asked about the sibling relationship on a 5-point Likert scale (1) Hardly at all 2) Not too much 3) Somewhat 4) Very much 5) Extremely much). Scale scores are derived by averaging the items.

8.1.8 Parenting

Child-parent relationship scale - CPRS-Short form (Pianta, 1995): This questionnaire consists of 15 questions with parent/caregiver reporting on perceived conflicts (8 items) and closeness (7 items) with their child. The respondent states their feelings and beliefs about their relationship with their child and about the child's behaviour towards them, by responding to statements on a 5 point likert scale (Definitely does not apply/Not really/Neutral, not sure/Applies somewhat/Definitely applies). Items are scored 1 (Definitely does not apply) to 5 (Definitely applies). The CPRS generates a separate score for the individual conflicts and closeness constructs. Scores for the individual conflicts (range 2 – 14) and closeness (range 1-15) items are added together to give a score of both conflicts with their child and also how close they perceive themselves to be to their child. This measure was designed for

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a normative population. However, it has also been used successfully with children with developmental disabilities.

Alabama parenting questionnaire (short form, adapted) (Frick, 1991): This questionnaire usually consists of 42 items, examining five subscales of parenting. We have selected two subscales, namely positive parenting (6 items) and inconsistent discipline (6 items). Parents/caregivers are asked about their parenting behaviours on a five-point likert scale (Never/Almost never/Sometimes/Often/Always). Items are scored from 1 (never) to 5 (always). All items in each scale are summed to obtain a total scale score. This measure was designed for a normative population. However, it has also been used successfully in examining parenting behaviours in children with developmental disabilities.

Parental activities/involvement index: This is a 5 item questionnaire to be completed by parent/caregivers. This questionnaire was constructed for the purposes of this study. Some of the questions have been used in national UK surveys (such as the Millennium Cohort Study) and some were developed specifically for this study. Where relevant, items were adapted to be more appropriate for families of children with intellectual disabilities. The questions ask about parent-child interactions for activities and parental involvement with their child. Answers are provided on a 6 point likert scale (Not at all/Less often than once or twice a month/Once or twice a week/Several times a week/Every day). The 6 point likert scale is the same as that used in the Millennium Cohort Study home learning environment index.

8.1.9 Child behavioural and emotional well-being

Strengths and difficulties questionnaire (SDQ) (parent version)(Goodman, 1997): The same 25-item behavioural screening questionnaire used with siblings, will also be used to ask parents/caregivers about the child with intellectual disabilities. This measure is described in the section, 8.1.7 Siblings, above.

Developmental behaviour checklist - parent report (DBC-P) (Einfeld & Tonge, 2002): This is a 96 item measure of behavioural and emotional disturbances in 4-18 year olds with intellectual or developmental disabilities. It is completed by the parent/caregiver and can give a total behaviour problem score as an overall measure of emotional and behavioural problems. It also gives a measure of disturbance across five sub-scales: disruptive/anti-social behaviour, self-absorbed behaviour, communication, anxiety and social relating disturbance. Items are scored on a 0, 1 or 2 rating scale (0 = Not true as far as you know, 1 = Somewhat or sometimes true, 2 = Very true or often true). The DBC is scored at 3 levels; 1) The Total Behaviour Problem Score (TBPS) is an overall measure of

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emotional and behavioural problems. The DBC can also detect clinically significant levels of overall emotional and behavioural disturbance (a score of 46 or greater). 2) The five subscales scores give a measure of disturbance including: Disruptive/Anti-social Behaviour, Self-absorbed, Communication Disturbance, Anxiety and Social Relating. 3) Additionally Individual behaviour items indicate the prevalence and severity of individual items.

Vineland adaptive behaviour scale second edition (VABS-II) Sparrow, Balla & Cicchetti (2005): This consists of a 15-20 min interview with parents, and measures a person's adaptive level of functioning. The VABS-II assesses the adaptive skills of an individual, whose age ranges from preschool to 18 years old. This semi-structured interview measure contains a range of 297 items that provide an assessment of adaptive behaviour across four domains: Socialization, Communication, Daily Living Skills, and Motor Skills. These adaptive skills items are arranged in developmental sequence and not all 297 questions are asked in an interview. Rather, the standard administration procedure is that the interviewer estimates an adaptive level and asks in detail about skill items in this range to arrive at an accurate estimate of a child's abilities. An overall composite score, and domain scores, can be derived with reference to age during typical development during which children can perform the task items. Four standardised scores in total will be derived for the present research: Adaptive Behaviour Composite, Communication Skills, Daily Living Skills, and Socialisation Skills.

9 Data analysis

The approaches to statistical analysis of the data will vary according to the research aims:

1. Descriptive statistics and one-sample t tests (and non-parametric alternatives where appropriate) will be used to report on the levels of well-being in mothers, fathers and siblings; and to compare well-being in these families to normative data or other UK samples. These analyses will enable us to establish whether family members experience increased levels of psychological problems compared to families of children without disabilities.
2. Well-being scores, relationship quality, and parenting attitudes and beliefs between mothers and fathers in the same families will be examined using multi-level modelling to take account of the nested nature of data from parents within the same family.
3. Correlation and regression approaches will be used to explore factors associated with the well-being of mothers, fathers and siblings. Final models will be tested using structural equation modelling.
4. Correlation and regression approaches will be used to explore factors associated with the well-being of the child with intellectual disability. Final models will be tested using structural equation modelling.

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5. For longitudinal analyses focused on factors associated with the well-being of the child with intellectual disability and family members over time, structural equation modelling approaches will be used.

10 Good clinical practice

10.1 Ethical conduct of the study

The study will be conducted in accordance with the ethical principles of the British Psychological Society. Approval will be obtained from the National Health Service Research Ethics Committee (NHS REC) prior to commencement of the study.

10.2 Investigator responsibilities

The Chief Investigator is responsible for the overall conduct of the study, compliance with the protocol and any protocol amendments. The responsibilities of the Chief Investigator may be delegated to an appropriate member of the research team.

10.3 Confidentiality

All participant information will be stored in a manner to maintain participant confidentiality. As participants will be required to input their personal contact information in the survey, we will extract this information from SurveyMonkey and store it in a separate database. Participants will be made aware during the consent procedure that we will be storing this information to contact them about further participation and link their responses if they participate in next wave of the research. If participants agree on the consent form, we will also use their contact information to keep them up-to-date with the findings of the research. They will also be made aware that the analysis or any associated publications will not identify participants by name.

All the research data will be stored electronically and password protected. The password and access to the research data, including participant information, will be restricted to the Chief Investigator and the research team. Study information will not be released without the written permission of the participant, except as necessary for monitoring auditing by the sponsor, sponsor's designee, regulatory authorities or the research ethics committee.

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The Chief Investigator and research team will not disclose, or use for any other purpose other than performance of the study, any data, raw record or other unpublished, confidential information disclosed to the research team for the purpose of the study. Prior written agreement from the sponsor, or the sponsor designee, will be obtained for the disclosure of any confidential information to other parties.

Participants will be assigned a randomised identification number to maintain their anonymity. We will publish reports and give presentations about the results of the study. However the participants will not be identified individually in any way as for analysis purposes responses will be pooled together.

Participants' names and addresses will not be able to be identified in any report or presentation about the study.

10.4 Data protection

The research team will comply with the requirements of the Data Protection Act 1998 with regard to the collection, storage; processing and disclosure of personal information will uphold the Act's core principles. Access to collated participant data will be restricted to the Chief Investigator and members of research team only.

Electronic data will be stored on firewalled University computers and password protected personal computers limited to the research team. Files will be password protected and only accessible to the researchers responsible for the running of the study and the Chief Investigator. All procedures for data storage, processing and management will be in compliance with the Data Protection Act 1998. All participants will be given a unique study number for the purpose of analysis. Data will only be stored on University managed computers. Personal information including full name and address will be safely secured electronically and password protected. Access to personal information will be restricted to the Chief Investigator and the research team. Information about consent and the matching of participants from the same family will be kept on a separate management database. For each participant their confidential information (such as name, address) will be extracted from survey monkey and they will individually be given a participant number. We will also obtain their consent preferences, stating whether they opt-in to: a) the telephone survey b) contact for Wave 2, and c) receiving updates about the study. Members of the research team can then consult this database when contacting participants.

11 Study conduct responsibilities

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11.1 Protocol amendments

Any changes in research activity except those necessary to remove an apparent immediate hazard to the participant must be reviewed and approved by the Chief Investigator. Amendments to the protocol must be submitted in writing for approval to relevant Research Ethics Committee(s) for approval prior to the participants being enrolled into an amended protocol.

11.2 Protocol violations and deviations

The Chief Investigator should not implement any deviation from the protocol without agreement from the relevant Research Ethics Committee(s), except when necessary to eliminate an immediate hazard to participants.

In the event that the Chief Investigator needs to deviate from the protocol, the nature of and the reasons for the deviation should be recorded. If this necessitated a subsequent protocol amendment, this should be submitted to the relevant Research Ethics Committee(s) for review and approval if appropriate.

11.3 Study record retention

Study records will be archived in line with standard operating procedures on archiving. All study data will be stored electronically for a minimum of five years after the end of the current study to allow for audit and inspection by regulatory authorities and will be readily available upon request. Retention of study data will also enable further analyses for potential future research and the possibility of a 3rd wave of the study.

11.4 End of study

The end of the current study is defined by an end date by which the second wave of data collection and initial analyses will have been completed. This date is 31 December 2019.

12 Reporting, publications and notifications of results

12.1 Authorship policy

Ownership of the data arising from this study resides with the University of Warwick. Various publications will be prepared for peer review on the basis of the study data, and PhD theses will also be written and submitted by current and future co-investigators.

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12.2 Publication and presentations

All publications and presentations relating to the study will be authorised by the Chief Investigator.

13 References

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4.4: 1000 Families Survey – Final Version (Primary Respondent)



Cerebra 1,000 Families study

Primary caregiver survey

Please enter the following details below so that your survey responses can be matched with any other respondents from your household.

1. Child with intellectual disability

First name of your child with intellectual disability:	
Surname of your child with intellectual disability:	

2. Your postcode

3. If your child lives at a different address to you, please enter your child's postcode

Contacting you about the telephone interview

4. I agree to participate in a telephone interview with a researcher (as described in the information sheet).

Please select <u>ONE</u>	✓
Yes (Please complete Questions 5,6 and 7)	
No (Please go to Question 8)	

5. Please enter the following details so you can be contacted by a member of the research team to arrange a time for the telephone interview.

First name:	
Surname:	

6. Telephone number

Home telephone number:	
Mobile telephone number:	

7. Email address

Contacting you in 2 years' time

8. I agree that the research team can contact me in 2 years' time to invite me to participate in the follow up study.

Please select <u>ONE</u>	✓
Yes (Please complete Questions 9, 10 and 11)	
No (Please go to Question 12)	

9. Please provide the following details so that we can contact you for the follow-up study in 2 years' time.

First name:	
Surname:	
Address Line 1:	
Address Line 2:	
City/Town:	
Postcode:	

10. Telephone number

Home telephone number:	
Mobile telephone number:	

11. Email address

Email updates

12. I wish to be kept up to date by email with the progress and findings of the study and other research about families of individuals with intellectual disability.

Please select <u>ONE</u>	✓
No	
Yes (Please provide your email address below)	

Questions about you and your child with Intellectual Disability

13. Is your child with intellectual disability male or female?

Please select <u>ONE</u>	✓
Male	
Female	

Questions about you

14. Please indicate your relationship to the child with intellectual disability.

Please select <u>ONE</u>	✓
Biological mother	
Biological father	
Adoptive mother	
Adoptive father	
Stepmother	
Stepfather	
Foster mother	
Foster father	
Grandmother	
Grandfather	

Other (please describe)

15. How do you identify your gender?

Please select <u>ONE</u>	✓
Male	
Female	
Trans	
Prefer not to answer	

16. How would you describe your ethnicity?

Please select <u>ONE</u>	✓
Asian/Asian British: Indian	
Asian/Asian British: Pakistani	
Asian/Asian British: Bangladeshi	
Asian/Asian British: Chinese	
Asian other (please describe below)	
<input type="text"/>	

Black/African/Black British: African	
Black/African/Black British: Caribbean	
Black other (please describe below)	
<input type="text"/>	

Mixed/multiple ethnic groups: White and Black Caribbean	
Mixed/multiple ethnic groups: White and Black African	

Mixed/multiple ethnic groups: White and Asian	
Mixed other (please describe below)	

Other Ethnic group: Arab	
Ethnic other (please describe)	

White: English/Welsh/Scottish/Northern Irish/British	
White: Irish	
White: Travelling community	
White: Other (Please describe below)	

Any other ethnic background (Please describe below)	

17. Please select the highest level of your educational qualifications

Please select <u>ONE</u>	✓
No qualifications	
Some GCSEs passes or equivalent	
5 or more GCSEs at A*-C or equivalent	
5 A/AS Levels or equivalent	
Higher Education but below degree level	
Degree (e.g. BA, BSC, MA)	
Don't know	

18. Please select one option which best describes your status

Please select <u>ONE</u>	✓
In a job and currently working for an employer	
On maternity/paternity /parental leave from a job	
Self-employed	
A Full time student	
Doing voluntary work	
Looking after home and family	
Unemployed	
Do something else (please specify below)	

19. How is your health in general?

Please select <u>ONE</u>	✓
Very good	
Good	
Fair	
Bad	
Very bad	

20. Do you have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled you over a period of time or is likely to affect you over a period of time?

Please select <u>ONE</u>	✓
No	
Yes	

Questions about your household

21. In total how many people currently live in your home (including yourself)?

	Adults	Children
Number		

Data from research with families with a family member with a disability has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether people with different levels of financial resources have different experiences.

22. What is your total weekly household income (after any deductions e.g. income tax), including income from paid work, pension, Social Services Benefits (e.g. Job Seekers Allowance, DLA, Carers' Allowance, Attendance Allowance, Tax Credits, Housing Benefits, Pension Credits) etc.?

Please select <u>ONE</u>	✓
£200 or less	
Between £200 and £300	
Between £300 and £400	
Between £400 and £500	
Between £500 and £600	
Between £600 and £700	
Between £700 and £800	
Between £800 and £900	
Over £1000	

23. How well would you say you [and your husband/wife/partner] are managing financially these days?

Would you say you are ...

Please select <u>ONE</u>	✓
living comfortably?	
doing alright?	
just about getting by?	
finding it quite difficult?	
finding it very difficult?	

24. Suppose you only had one week to raise £2000 for an emergency, which of the following best describes how hard it would be for you to get that money?

Please select ONE	✓
I could easily raise the money	
I could raise the money, but it would involve some sacrifices (e.g. reduced spending, selling a possession)	
I would have to do something drastic to raise the money (e.g. selling an important possession)	
I don't think I could raise the money	

Questions about your child with intellectual disability

We would now like to ask you some questions about your child with intellectual disability who you named earlier in the survey.

25. The date of birth of your child with intellectual disability

DD MM YY

26. Please select which of the conditions below professionals have diagnosed in relation to your child with intellectual disability (select ALL that apply)

Select <u>ALL</u> that apply	✓
Learning disability/learning difficulty	
Autism/Autistic Spectrum Disorder /Autistic Spectrum Condition/Asperger's Syndrome	
Down syndrome	
Global Developmental delay	
Cerebral palsy	
Other genetic syndrome/diagnosis (please describe below)	
<input type="text"/>	

27. Please state if your child with intellectual disability has:

Please select ONE	✓
A mild/moderate intellectual disability Children with a mild to moderate intellectual disability can typically communicate and look after themselves well, but may take a bit longer to learn new skills compared to other children of the same age.	
A Severe/profound intellectual disability Children with a severe to profound intellectual disability are likely to have complex and multiple difficulties which require extensive support to learn and carry out daily activities.	

28. Does your child with intellectual disability have a visual impairment?

Please select ONE	✓
Yes	
No	

29. Does your child with intellectual disability have a hearing impairment?

Please select <u>ONE</u>	✓
Yes	
No	

30. Does your child with intellectual disability currently have epileptic seizures?

Please select <u>ONE</u>	✓
Yes (Please go to Question 32)	
No (Please go to Question 31)	

31. Has your child with intellectual disability ever had an epileptic seizure in the past?

Please select <u>ONE</u>	✓
Yes	
No	

32. Does your child with intellectual disability have any mobility problems?

Please select <u>ONE</u>	✓
Yes	
No	

33. Does your child with intellectual disability have any other physical health problems?

Please select <u>ONE</u>	✓
Yes	
No	

34. Does your child with intellectual disability normally

Please select <u>ONE</u>	✓
Live with you full-time? (Please go to Question 36)	
Live with you part-time? (Please go to Question 35)	

35. Please state the approximate number of hours that your child lives with you on a weekly basis

36. What type of school does your child with intellectual disability usually attend?

Please select <u>ONE</u>	✓
Mainstream school	
Mainstream school in either a special unit or resourced Special Educational Needs (SEN) provision	
Special school	
Home schooled	
Not currently in school	

Questions about your experiences

The following statements ask about your experiences of having a child with intellectual disability.

37. Please respond to all questions by selecting the response which best describes how you feel about each statement.

Please select <u>ONE</u> answer per statement	Strongly agree	Agree	Neither agree or disagree	Disagree	Strongly disagree
Since having this child I have grown as a person					
Having this child has helped me learn new things/skills					
Raising this child helps put my life into perspective					
Since having this child, my family has become closer to one another					
Since having this child, my family has become more tolerant and accepting					
Since having this child I have become more determined to face up to challenges					
Since having this child I have a greater understanding of other people					

We would now like to ask you about the time you have to do leisure or social activities.

38. Has the ability to spend time doing leisure or social activities been affected by the assistance you give to your child with intellectual disability in any of the ways described below? (For all responses, this should be as a result of the caring and not for other reasons). Please select ANY that apply.

Please select <u>ANY</u> that apply	✓
Unable to socialise or take part in social or leisure activities at all (due to caring responsibilities)	
Reduced time with spouse or partner	
Reduced time with other family members	
Reduced time with friends	
Difficulties making new friends	
Reduced time spent doing sport or physical activity	
Reduced time spent doing pastime or hobby	

The following questions ask about how you have been feeling during the past 30 days.

39. For each question, please click the number that best describes how often you had this feeling. During the past 30 days, about how often do you feel

Please select <u>ONE</u> answer per statement	All of the time	Most of the time	Some of the time	A little of the time	None of the time
...nervous?					
...hopeless?					
...restless or fidgety?					
...so depressed that nothing could cheer you up?					
...that everything was an effort?					
...worthless?					

We are now going to ask you about your satisfaction with life.

40. Here is a scale from 1-10 where '1' means that you are completely dissatisfied and '10' means that you are completely satisfied. All things considered, please could circle the number which corresponds with how satisfied or dissatisfied you are about the way your life has turned out so far.

1	2	3	4	5	6	7	8	9	10
---	---	---	---	---	---	---	---	---	----

41. What is your current marital status?

Please select <u>ONE</u>	<input checked="" type="checkbox"/>
Married and living with spouse/civil partner (Please answer questions 42 and 43)	<input type="checkbox"/>
Living with partner (Please answer questions 42 and 43)	<input type="checkbox"/>
Divorced/Separated/Single/Widowed/Not currently living with partner (Please go to question 44)	<input type="checkbox"/>

We would now like to ask you some questions about your relationship with a spouse/partner.

42. How often do you and your [husband/wife/partner] disagree over issues relating to your child with intellectual disability?

Please select <u>ONE</u>	<input checked="" type="checkbox"/>
Never	<input type="checkbox"/>
Less than once a week	<input type="checkbox"/>
Once a week	<input type="checkbox"/>
Several times a week	<input type="checkbox"/>
Once a day	<input type="checkbox"/>
More than once a day	<input type="checkbox"/>
Can't say	<input type="checkbox"/>

43. Here is a scale from 1-7 where '1' means that you are very unhappy and '7' means that you are very happy. Please circle the number which best describes how happy or unhappy you are with your relationship with your spouse/partner, all things considered?

1	2	3	4	5	6	7	8
Very unhappy						Very happy	Can't say

Questions about your family

We would now like to ask you about how satisfied you are with family life.

44. Please read the following statements and select what best applies to you.

Please select <u>ONE</u> answer per statement	Almost always	Some of the time	Hardly ever
I am satisfied that I can turn to my family for help when something is troubling me	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I am satisfied with the way my family talks over things with me and shares problems with me	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I am satisfied that my family accepts and supports my wishes to take on new activities or directions	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I am satisfied with the way my family and I share time together	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Your child's strengths and difficulties

We would now like to ask about the strengths and difficulties of your **child with intellectual disability**. If there are any items that do not apply to your child then please tick 'Not True'.

45. Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the **last six months**.

	Not true	Somewhat true	Certainly true
Considerate of other people's feelings			
Restless, overactive, cannot stay still for long			
Often complains of headaches, stomach-aches or sickness			
Shares readily with other children (treats, toys, pencils etc.)			
Often has temper tantrums or hot tempers			
Rather solitary, tends to play alone			
Generally obedient, usually does what adults request			
Many worries, often seems worried			
Helpful if someone is hurt, upset or feeling ill			
Constantly fidgeting or squirming			
Has at least one good friend			
Often fights with other children or bullies them			
Often unhappy, down-hearted or tearful			
Generally liked by other children			
Easily distracted, concentration wanders			
Nervous or clingy in new situations, easily loses confidence			
Kind to younger children			
Often lies or cheats			
Picked on or bullied by other children			
Often volunteers to help others (parents, teachers, other children)			
Thinks things out before acting			
Steals things from home, school or elsewhere			
Gets on better with adults than with other children			
Many fears, easily scared			
Sees tasks through to the end, good attention span			

Relationship with your child with intellectual disability

We would now like to ask you about your relationship with your child with intellectual disability.

46. Please reflect on the degree to which each of the following statements currently applies to your relationship with your child with intellectual disability. Using the scale below, tick one appropriate answer for each item.

Please select <u>ONE</u> answer per statement	Definitely does not apply	Not really	Neutral, not sure	Applies somewhat	Definitely applies
I share an affectionate relationship with this child					
This child and I always seem to be struggling with each other					
If upset, this child will seek comfort from me					
This child is uncomfortable with physical affection or touch from me					
This child values his/her relationship with me					
When I praise this child, he/she beams with pride					
This child spontaneously shares information about himself/herself					
This child easily becomes angry with me					
It is easy to be in tune with what this child is feeling					
This child remains angry or is resistant after being disciplined					
Dealing with this child drains my energy					
When this child is in a bad mood, I know we're in for a long difficult day					
This child's feelings towards me can be unpredictable or change suddenly					
This child is sneaky or manipulative with me					
This child openly shares his/her feelings and experiences with me					

Parenting your child with intellectual disability

We would now like to ask you some questions about your approach to parenting your child with intellectual disability.

47. The following are a number of statements about your approach to parenting your child with intellectual disability. Please rate each item as to how often it typically occurs in your home.

Please select <u>ONE</u> answer per statement	Never	Almost Never	Sometimes	Often	Always
You let your child know when he/she is doing a good job with something					
You threaten to punish your child and then do not actually punish him/her					
You reward or give something extra to your child for obeying you or behaving well					
Your child talks you out of being punished after he/she has done something wrong					
You feel that getting your child to obey you is more trouble than it's worth					
You compliment your child when he/she does something well					
You praise your child if he/she behaves well					
You let your child out of a punishment early (e.g., lift restrictions earlier than you originally said.)					
You hug or kiss your child when he/she has done something well					
The punishment you give your child depends on your mood					
Your child is not punished when he/she has done something wrong					
You tell your child that you like it when he/she helps around the house					

We would now like to ask you about how often you do particular activities with your child with intellectual disability.

48. The following are a number of statements about you and your child with intellectual disability. Please rate each item as to how often it has typically occurred during the past six months.

Please select <u>ONE</u> answer per statement	Not at all	Less often than once or twice a month	Once or twice a month	Several times a week	Everyday
How often do you read or share a story with your child?					
How often do you and your child sing together?					
How often do you and your child play a game together?					
How often do you and your child go out together for enjoyment? (rather than as a chore e.g. appointments/ food shopping etc.)					
How often do you and your child watch TV together?					

49. Does your child with intellectual disability have at least one sibling between the ages of 4 to 15?

Please select <u>ONE</u>	✓
Yes (Please go to Question 50)	
No (Please go to the end of the survey)	

Sibling strengths and difficulties

We would now like to ask about the strengths and difficulties of one of the siblings of your child with intellectual disability. If there is more than one sibling between the ages of 4 to 15 please choose the sibling closest in age to your child with intellectual disability.

50. Age of sibling in years and months

Years Months

51. Sibling gender

Please select ONE	✓
Male	
Female	

52. Does this sibling have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled them over a period of time or is likely to affect them over a period of time?

Please select ONE	✓
No	
Yes (Please also answer question 54)	

53. Does this sibling live in the same household as your child with intellectual disability?

Please select ONE	✓
All of the time	
Some of the time	
None of the time	

54. Please give details of the sibling's longstanding illness, disability or infirmity.

We would now like to ask about the strengths and difficulties of the **sibling of your child** with intellectual disability. If there are any items that do not apply to your child then please select 'Not True'.

55. Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the **last six months**.

	Not true	Somewhat true	Certainly true
Considerate of other people's feelings			
Restless, overactive, cannot stay still for long			
Often complains of headaches, stomach-aches or sickness			
Shares readily with other children (treats, toys, pencils etc.)			
Often has temper tantrums or hot tempers			
Rather solitary, tends to play alone			
Generally obedient, usually does what adults request			
Many worries, often seems worried			
Helpful if someone is hurt, upset or feeling ill			
Constantly fidgeting or squirming			
Has at least one good friend			
Often fights with other children or bullies them			
Often unhappy, down-hearted or tearful			
Generally liked by other children			
Easily distracted, concentration wanders			
Nervous or clingy in new situations, easily loses confidence			
Kind to younger children			
Often lies or cheats			
Picked on or bullied by other children			
Often volunteers to help others (parents, teachers, other children)			
Thinks things out before acting			
Steals things from home, school or elsewhere			
Gets on better with adults than with other children			
Many fears, easily scared			
Sees tasks through to the end, good attention span			

56. We would now like you to tell us about the relationship between your child with intellectual disability and their sibling you have just told us about.

Please select <u>ONE</u> answer per statement	Hardly at all	Not too much	Somewhat	Very much	Extremely much
How much do the sibling and the child tell each other everything?					
How much do the sibling and the child share secrets and private feelings?					
How much do the sibling and the child go places and do things together?					
Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do the sibling and the child play around and have fun with each other?					
Some siblings care about each other a lot while other siblings don't care about each other that much. How much do the sibling and the child care about each other?					
How much do the sibling and the child love each other?					
How much do the sibling and the child disagree and quarrel with each other?					
How much do the sibling and the child get mad and get in arguments with each other?					
How much do the sibling and the child insult and call each other names?					
How much are the sibling and the child mean to each other?					

End of survey

Thank you for completing the 1,000 Families survey. **Please return the survey with your consent form using the pre-post envelope in your pack.**

If you wish to forward this survey onto other parents, this would be greatly appreciated. The link to the online survey is www.surveymonkey.com/r/1000families or alternatively a paper copy can be requested by emailing: familyresearch@warwick.ac.uk.

For updates on this study and other topics related to families of children with intellectual disability please visit our Facebook and Twitter pages.



www.facebook.com/FamilyRG1



[@Family_RG1](https://twitter.com/Family_RG1)

4.5: Participant (Primary Respondent) Information Sheet – Final Version



1,000 Families study

Primary caregiver study information

Thank you for taking part in the 1,000 families study. Your participation in this study will make a valuable contribution to UK based family and disability research.

We estimate that the survey will take around 20 minutes for you to complete.

On the next page there is information about the study as required and approved by the NHS ethics committee. Please keep a copy of this information to read at a time of your convenience.

Primary caregiver Information sheet

We would like to invite you to take part in our research study. Before you decide to take part it is important for you to understand what the research would involve. Please take time to read the following information carefully. If there is anything that is unclear, or if you would like more information please contact us using the details provided at the end.

Purpose of the study

The aim of this study is to understand the experiences of family members living in the UK who care for a child with an intellectual disability (often called learning disability or learning difficulties in the UK). We wish to capture the experiences of parents/parental caregivers and any siblings. We will be recruiting a large number of families (at least 1,000 Families) to take part in this research. We would also like to follow families over time, because it is important to understand how families change over time and adjust in different ways to having a child with intellectual disability.

Who can take part?

We are inviting primary parental caregivers of children with intellectual disability (learning disability or learning difficulties) between the ages of 4 and 15 years 11 months of age to take part in this research. Primary parental caregivers might not be the child's mother, but would be the adult who cares for the child with intellectual disability for most of the time. Mothers may be biological, adoptive, or foster mothers. Your child with intellectual disability might also have other diagnosed conditions such as autism, Down syndrome, or other genetic syndromes. Your family must currently live somewhere in the UK. This research focuses on families whose child with intellectual disability lives with them for the majority of the time (more than half of a typical week). If your child with intellectual disability lives outside of the family home for the majority of the week (e.g., in a residential school placement) then this research study is not for you.

Why have I been invited to take part in the research?

You have been invited to take part in this research because you are a parent or parental caregiver of a child with intellectual disability aged between 4 and 15 years 11 months of age and living in the UK.

What will happen if I take part?

Once you have read this study information, you will be asked to read some

statements and to indicate your agreement to each one. We need to check that you agree to participate in the research. The online survey then includes questions asking about you and your family, your experiences as a parent, about your child with intellectual disability, and also about a sibling in the family if there is one. We expect the online survey to take about 20 minutes to complete in total.

If you agree, a researcher will then telephone you to ask you some additional questions about your child with intellectual disability. These questions are much easier to ask in a telephone interview, and focus on understanding the skills that your child with intellectual disability has and on some of the difficulties that they may face. This telephone interview usually takes about 40-60 minutes. We can schedule the interview for a time that suits you, and it is easy to re-arrange a time if the first time organised ends up being inconvenient for you.

If you agree, we will send you updates about this research project and other research that we are carrying out about the experiences of families of individuals with intellectual disability.

We plan to follow up with 1,000 Families Study families in about two years' time. If you agree, we will contact you again then to ask if you would like to take part in this follow-up research. At that stage, we would ask you to complete the online survey and the telephone interview again.

Do I have to take part in the research?

You are under no obligation to participate in the research, your involvement is voluntary. You can decide to complete the online survey and not the telephone interview. You can also complete the online survey and telephone interview, but not give us permission to invite you to take part again in two years' time.

If you do agree to participate and then decide you no longer want to take part, you are free to withdraw. You do not have to provide a reason for wanting to withdraw from the study. You would have the option for any data you have provided up until your withdrawal to be removed from the study and destroyed.

You are able to withdraw yourself from the study even if another parent/caregiver from your household has also participated in the study.

Apart from the questions checking that you are happy to take part in the various parts of the study, you do not have to answer any questions that you do not wish to answer.

Are there any possible benefits and risks of taking part in the study?

Involvement in this research provides an opportunity to share you and your family's

experiences of raising a child with an intellectual disability. The information you provide will help us to understand more about families like yours, to share this information widely, and to inform ways to better support families.

We do not anticipate any risks to taking part in this research. The questions that we are asking have been used in several research studies before, and we have removed any that have tended to cause family members distress. However, it is possible that you will find some of the questions to be upsetting because we do ask about your well-being and some of the difficulties faced by you, your family and your child with intellectual disability. We do also ask about positive experiences. If you are upset by any of the questions, you do not have to respond to them and you are under no obligation to continue with the survey or interview.

If any of the survey or interview questions make you concerned for yourself or another family member's well-being, we recommend that you make contact with your General Practitioner (GP) or one of the helplines listed below:

Mencap: 0808 808 1111, Contact a Family: 01332 557 975, KIDS: 0207 359 3635

How will the information you give to us be looked after?

All information that you provide as a part of this study will remain confidential, and we will store the information securely (in locked cabinets, or secure password protected computers) in an anonymised form. Access is restricted to the research team.

We will publish reports and give presentations about the results of the study. However, you will not be identified individually in any way as your responses will be pooled together with other participants and you will be assigned an anonymised study number. You and your family will not be able to be identified in any report or presentation about the study.

There are circumstances in which we would not be able to keep confidential something that you say. If you mention during the telephone interview any information that suggests someone in your family, or you, is at risk of harm or has been subject to abuse, the researcher would have a duty to report this information to the appropriate authorities.

We will keep your personal data for three years so that we can invite you to take part in the research study again in approximately two years' time. At that time, we will check again if we can approach you again in the future. Your anonymised responses to the survey and interview questions will be archived so that researchers in the future can carry out additional analysis of the data from the 1,000 Families Study. These researchers would not have access to your personal information and would have to agree to abide by appropriate ethical principles to do any new research.

Who is organising and funding the research?

The University of Warwick is responsible for this research. The research has received funding from the charity Cerebra and through the Economic and Social Research Council Doctoral Training Centre at the University of Warwick.

Who has reviewed the study?

This study has been reviewed and given a favourable opinion by a NHS Research Ethics Committee. A Research Ethics Committee is a group of independent people who review research to protect the dignity, rights, safety, and well-being of participants and researchers.

Further information and contact details

If you would like to ask questions before deciding whether to participate, please contact a member of the research team (Tel: 02476 524 139, Email: familyresearch@warwick.ac.uk).

If you are unhappy about any aspect of this research study, please contact the Chief Investigator Richard Hastings by mail, email or telephone (CEDAR, University of Warwick, Coventry, CV4 7AL; R.Hastings@warwick.ac.uk; 02476 522 197).

*If you would like to discuss the research, or ask any questions, with **someone who is not a part of the study team**, please contact ; Ms. Samantha Flynn (Email: S.Flynn.1@warwick.ac.uk; 02476 524 139).*

This study is covered by the University of Warwick's insurance and indemnity cover.

Any complaint about the way that you have been dealt with during the study, or any possible harm that you might have suffered, should be directed to: : the Head of Research Governance, Research & Impact Services, University House, University of Warwick, Coventry, CV4 8UW. Email-researchgovernance@warwick.ac.uk; telephone : 024 76 522746

4.6: Participant (Primary Respondent) Consent Form – Final Version



1,000 Families study

Primary caregiver consent form

Please send this with your survey in the pre-post envelope.

*Please read carefully the initial statements below. If you agree with these statements then tick in the corresponding box. **Unfortunately if you do not consent to all of the statements we cannot use your survey responses.***

If you agree with these statements then tick the corresponding box.

- I confirm that I have read and understood the information provided for the 1,000 Families study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.
- I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without my rights being affected.
- I understand that relevant sections of my data collected during the study, may be looked at by individuals from regulatory authorities responsible for investigating research fraud. I give permission for individuals from these regulatory authorities to have access to my records if research fraud in this study was to be investigated.
- I agree to participate in the survey.

4.7: Example Advertisement and Recruitment Documents

Cerebra 1,000 Families Study

If you are the mother, father or caregiver of children with a learning disability, this is your opportunity to inform research and policy for coming decades!

We understand that families play a vital role in their children's lives. By listening to what families tell us we are able to help families see past barriers and discover a better life together.

Together with the Cerebra Family Research Group at the University of Warwick we are exploring the experiences of family members who live with a child with a learning disability aged **4 to 11 years**. You can also take part if your child has a learning disability and other conditions such as Down syndrome, autism, or other genetic syndromes.

The study involves taking a short survey and we are really keen to hear from *at least* one person in the family. We would like to hear from fathers as well as mothers.

We want to make this the largest study ever of families of children with learning disability in the UK. The 1,000 families study will further understanding of what it is like to raise a child with a learning disability and will help shape future information and support for families and children – so please get involved.

To find out more about the study and to complete the online survey please [visit the website](#). If you prefer you can request a [paper copy of the survey](#).

If you have any questions please email: familyresearch@warwick.ac.uk

Thank you for taking the time to consider taking part in this important family research project. We would be very grateful if you could forward this email on to anyone else you think might be interested and help us to reach 1,000 families!

We're the charity dedicated to helping families with children with brain conditions discover a better life together. It's an incredibly rewarding journey for everyone involved. Why not be part of it? You never know what we'll discover together

Best wishes

THE CEREBRA FAMILY RESEARCH GROUP

CEDAR
 University of Warwick
 Coventry CV4 7AL, United Kingdom
 Tel: +44 (0) 24 7652 3952
 Fax: +44 (0) 24 7652 4472
 Email: familyresearch@warwick.ac.uk



THE CEREBRA FAMILY RESEARCH GROUP

UNIVERSITY OF WARWICK

1,000 Families Study

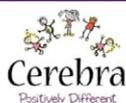
Are you the parent or parental caregiver of a child aged ~~4-11 years~~ 4 years - 15 years and 11 months that has a learning disability? If so, please take part in our new 'Cerebra 1000 Families study.' Your child might have other conditions - such as Autism, or any one of a number of genetic syndromes. As long as your child also has a learning disability, we would be very keen to hear from you.

The Cerebra Family Research Group, at the University of Warwick, in collaboration with the research charity Cerebra, wishes to explore the experiences of families who live with a child with a learning disability in the UK. We want to make this the largest study ever of families of children with learning disability in the UK.

This research will further our understanding of what it is like to raise a child with a learning disability in the UK and will help shape future support for families and children.

To find out more about the research, please follow the link below to our survey.

--Surveymonkey link --



1,000 Families study

Are you the mother, father, or caregiver of a child aged between 4 and 15 years, 11 months who has a learning disability?

If so, please take part in our new 'Cerebra 1000 Families study.' Your child might have other conditions - such as Autism, or any one of a number of genetic syndromes. As long as your child also has a learning disability, we would be very keen to hear from you.

The Cerebra Family Research Group at the University of Warwick, in collaboration with the research charity Cerebra, wishes to explore the experiences of **family members who live with a child with a learning disability in the UK.**

We are really keen to learn about the experiences of **at least one person** in the family. Most research studies only ask one person in the household to report on their family, however if **more than one family member** participates (especially a mother and a father) we will be able to explore whether family members have similar or different experiences.

We want to make this the **largest** study ever of families of children with a learning disability in the UK



www.facebook.com/FamilyRG1



@Family_RG1

This research will **further our understanding** of what it is like to raise a child with a learning disability in the UK and will **help shape future support** for families and children.

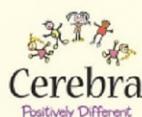
To take part please go to:

www.surveymonkey.com/r/1000families

If you have any questions or would like to know more about the study, please contact the research team by email: familyresearch@warwick.ac.uk or telephone: **02476 524 139**

Cerebra 1,000 families study

What is it like raising a child with an intellectual (learning) disability in the UK? We want to hear from you about your experiences.



To take part visit
www.surveymonkey.com/r/1000families



Take part in our survey

Our study will raise awareness of what it is like to raise a child with an intellectual (learning) disability in the UK and will shape decades of research and support for families.

For more information or to request a paper copy of the survey contact us at familyresearch@warwick.ac.uk



@family_RG1



facebook.com/familyRG1

Who can take part?



You can be a mother, father or family member of a child who is aged between 4 and 11 years AND has an intellectual (learning) disability (your child may have other conditions such as autism, or any one of a number of genetic syndromes)

What will it involve?



Online or paper survey (approx. 20 mins)

Telephone interview (optional and usually with the primary caregiver)

THE CEREBRA FAMILY RESEARCH GROUP

CEDAR
University of Warwick
Coventry CV4 7AL, United Kingdom
Tel: +44 (0) 24 7652 3952
Fax: +44 (0) 24 7652 4472
Email: familyresearch@warwick.ac.uk



THE UNIVERSITY OF
WARWICK

THE CEREBRA FAMILY RESEARCH GROUP
UNIVERSITY OF WARWICK

1,000 Families Study

Are you the parent or parental caregiver of a child aged 4-11 years that has a learning disability? If so, please take part in our new 'Cerebra 1000 Families study.' Your child might have other conditions - such as Autism, or any one of a number of genetic syndromes. As long as your child also has a learning disability, we would be very keen to hear from you.

The Cerebra Family Research Group, at the University of Warwick, in collaboration with the research charity Cerebra, wishes to explore the experiences of families who live with a child with a learning disability in the UK. We want to make this the largest study ever of families of children with learning disability in the UK.

This research will further our understanding of what it is like to raise a child with a learning disability in the UK and will help shape future support for families and children.

To find out more about the research, please follow the link below to our survey.

--Surveymonkey link --

4.8: Response to Ethical Review Comments

Cerebra Family Research Group

CEDAR
 University of Warwick
 Coventry CV4 7AL, United Kingdom
 Tel: +44 (0) 24 7652 3952
 Fax: +44 (0) 24 7652 4472
 Email: familyresearch@warwick.ac.uk



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WARWICK

Response to ethics committee

Dear Ms Gregory

Thank you for your letter of 27 August with the committee's decision on our research ethics application. We have acted on the committee's requested changes as detailed below.

1. The Committee require reference to siblings name and date of birth to be removed from the 'The Cerebra 1000 Families Survey' document. The age range for siblings should also be corrected from up to 18 years to up to 16 years of age. It is acceptable to ask the sex and age of the sibling but not the name and date of birth.

We have removed the request for the sibling's name and date of birth. Instead, we have asked for the sibling's current age in years and months. We have also made clear that the siblings must be at age 4 and under 16 years of age.

2. The Committee require the following amendments to be made to the 'The Cerebra 1000 Families Survey' document. 'Mother' should be replaced with 'primary carer', ethnicity should be listed in alphabetical order, and 'Gypsy or Irish Traveller' should be replaced with 'Travelling Community'.

We could find no reference to "mother" in this survey document. However, we did realise that we needed to ask caregivers about their relationship to the child and so have added a question to cover this (new question 3). We have also made the changes to the ethnicity question as requested.

3. The Committee note that the Vineland Adaptive Behaviour Scale was validated for use in face to face interviews but the interviews in this particular study are taking place by telephone. The Committee request the applicant's comments.

The VABS requires an interview with a carer but there is no requirement to be in sight of the carer when conducting the interview. We have carried out several hundred VABS interviews over the telephone. Carers have also preferred this method since we do not have to make appointments to meet them in their homes. We have found carers to be more relaxed over the telephone because they are comfortable to request a revised call time if they are busy. Subsequently, we are confident that the data obtained in a telephone interview are at least as valid as data obtained face-to-face, and perhaps more so.

4. The Committee require assurance that appropriate instructions will be supplied to participants completing the online survey that responses to questions may be left blank if the participant chooses.

This instruction was already included in both information sheets – to be placed at the beginning of the online survey - and remains in place.

Although not explicitly requested to do so, in response to the discussion at the committee we have included further reassurance to parents at the beginning of the Strengths and Difficulties Questionnaire when being completed on the child with learning disability. We

cannot change the SDQ instructions due to copyright restrictions when presenting this online. However, we added a statement to suggest parents might respond "never true" if they feel that any item does not apply to their child. They are still welcome, as with all questions, to not respond.

5. The following changes should be made to the participant information sheet:

- a) Provide an independent source of advice at the end of the participant information sheet should the participant wish to discuss the research with someone unconnected with the study.

We have added Charlotte Rowe's name and email contact. Charlotte is a CEDAR researcher who is not a part of the 1000 Families study team. We also have a space for the telephone number. These are in the process of change at the present, and we will add the correct number before data collection starts.

6. The following changes should be made to the consent form:

- a) Include the standard paragraph regarding access to data and regulatory authorities.

We have added a paragraph to both consent forms to be clear that regulatory bodies investigating research fraud may require access to the participants' data.

Although not explicitly asked to do so, we have also updated the University of Warwick insurance document to the one for the current year to 31 July 2016.

We hope that these changes meet with the committee's requirements and look forward to hearing from you.

Yours sincerely

Richard Hastings

Professor, and Cerebra Chair of Family Research

4.9: Approval Letter from Ethics Committee



Health Research Authority
NRES Committee West Midlands - South Birmingham

Royal Standard Place
 Nottingham
 NG1 6FS

Tel: 0115 883 9428

11 September 2015

Professor Richard Hastings
 CEDAR (Centre for Educational Development Appraisal and Research)
 University of Warwick
 Coventry UK
 CV47AL

Dear Professor Hastings

Study title:	The 1,000 families study: Well-being in families of children with intellectual disability
REC reference:	15/WM/0267
IRAS project ID:	169882

Thank you for your letter of 07 September 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Assistant, Nicola Kohut, nrescommittee.westmidlands-southbirmingham@nhs.net. Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Copies of advertisement materials for research participants [Project short advertisement text]	1	13 July 2015
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Warwick University insurance 15-16]		
IRAS Checklist XML [Checklist_07092015]		07 September 2015
Letter from sponsor [Confirmation of Sponsorship]		
Other [CV Mikeda Jess]		
Other [CV Jane Margetson]		
Other [Elizabeth Halstead CV]		
Other [Response to initial ethics opinion]		
Participant consent form [Primary parental caregiver consent form]	1.1	03 September 2015
Participant consent form [Secondary Parental Caregiver consent form]	1.1	03 September 2015
Participant information sheet (PIS) [Primary caregiver information sheet]	1.1	03 September 2015
Participant information sheet (PIS) [Secondary caregiver information sheet]	1.1	03 September 2015
REC Application Form [REC_Form_14072015]		14 July 2015
Research protocol or project proposal [1000 Families study protocol]	1	09 July 2015
Summary CV for Chief Investigator (CI) [Richard Hastings CV]		
Summary CV for student [CV Emma Langley]		
Summary CV for supervisor (student research) [CV Vaso Totsika]		
Validated questionnaire [Developmental Behaviour Checklist (DBC)]		
Validated questionnaire [Online survey items and questionnaires]	1.1	03 September 2015

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at

<http://www.hra.nhs.uk/hra-training/>

15/WM/0267

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely



Professor Simon Bowman
Chair

Email: nrescommittee.westmidlands-southbirmingham@nhs.net

Enclosures: After ethical review – guidance for researchers

Copy to: Mrs Jane Prewett

4.10: Notice of Ethics Amendment

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

Please enter a short title for this project (maximum 70 characters)
The 1,000 Families Study

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located?(Tick all that apply)

- England
- Scotland

<input checked="" type="checkbox"/> Wales <input checked="" type="checkbox"/> Northern Ireland 3a. In which country of the UK will the lead NHS R&D office be located: <input type="radio"/> England <input type="radio"/> Scotland <input type="radio"/> Wales <input type="radio"/> Northern Ireland <input checked="" type="radio"/> This study does not involve the NHS
4. Which applications do you require? <input type="checkbox"/> NHS/HSC Research and Development offices <input type="checkbox"/> Social Care Research Ethics Committee <input checked="" type="checkbox"/> Research Ethics Committee <input type="checkbox"/> Confidentiality Advisory Group (CAG) <input type="checkbox"/> National Offender Management Service (NOMS) (Prisons & Probation) <p><i>For NHS/HSC R&D Offices in Northern Ireland, Scotland and Wales the CI must create NHS/HSC Site Specific Information forms, for each site, in addition to the study wide forms, and transfer them to the PIs or local collaborators.</i></p> <p><i>For participating NHS organisations in England different arrangements apply for the provision of site specific information. Refer to IRAS Help for more information.</i></p>
5. Will any research sites in this study be NHS organisations? <input type="radio"/> Yes <input checked="" type="radio"/> No
6. Do you plan to include any participants who are children? <input type="radio"/> Yes <input checked="" type="radio"/> No
7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves? <input type="radio"/> Yes <input checked="" type="radio"/> No <p><i>Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.</i></p>
8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales? <input type="radio"/> Yes <input checked="" type="radio"/> No
9. Is the study or any part of it being undertaken as an educational project? <input checked="" type="radio"/> Yes <input type="radio"/> No

Notice of Amendment

IRAS Version 5.4.0

Please describe briefly the involvement of the student(s):
Three PhD students will work on data collection for this survey. Each will be examining and reporting data from a different area of the survey, with their own research questions, to contribute towards their PhD theses.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

Yes No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

Yes No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

Yes No

4.11: Notice of Substantial Amendment

NOTICE OF SUBSTANTIAL AMENDMENT

Please use this form to notify the main REC of substantial amendments to all research other than clinical trials of investigational medicinal products (CTIMPs).
The form should be completed by the Chief Investigator using language comprehensible to a lay person.

Details of Chief Investigator:

	Title	Forename/Initials	Surname
	Professor	Richard	Hastings
Work Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
PostCode	CV47AL		
Email	[REDACTED]k		
Telephone	[REDACTED]		
Fax			

For guidance on this section of the form refer to the guidance

Full title of study:	The 1,000 families study: Well-being in families of children with intellectual disability
Lead sponsor:	University of Warwick
Name of REC:	West Midlands - South Birmingham
REC reference number:	15/WM/0267

Additional reference number(s):

Ref.Number	Description	Reference Number
	Sponsor reference number	REGO-2015-1604

Name of lead R&D office:

Date study commenced:	26th November 2015
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Protocol reference (if applicable), current version and date:

Amendment number and date:	Version 1 (13/01/2017)
-----------------------------------	------------------------

Type of amendment

(a) Amendment to information previously given in IRAS

Yes No

If yes, please refer to relevant sections of IRAS in the "summary of changes" below.

A17-1 inclusion criteria upper age for children increased to 15 years 11 months

(b) Amendment to the protocol

Yes No

If yes, please submit either the revised protocol with a new version number and date, highlighting changes in bold, or a document listing the changes and giving both the previous and revised text.

Revised protocol attached with updates to change upper age for inclusion. We have also taken the opportunity to update research team and other contact information. Amended protocol showing all tracked changes attached.

(c) Amendment to the information sheet(s) and consent form(s) for participants, or to any other supporting documentation for the study

Yes No

If yes, please submit all revised documents with new version numbers and dates, highlighting new text in bold.

Upper age for inclusion amended, and updated information about an independent research contact for carers wishing to ask questions about the study.

Is this a modified version of an amendment previously notified and not approved?

Yes No

Summary of changes

Briefly summarise the main changes proposed in this amendment. Explain the purpose of the changes and their significance for the study.

If this is a modified amendment, please explain how the modifications address the concerns raised previously by the ethics committee.

If the amendment significantly alters the research design or methodology, or could otherwise affect the scientific value of the study, supporting scientific information should be given (or enclosed separately). Indicate whether or not additional scientific critique has been obtained.

At present, we are recruiting caregivers raising children with Intellectual Disability (ID) who are between the ages of 4 years to 11 years and 11 months. We would like to change our inclusion criteria to extend the maximum age of the child with ID at recruitment 15 years and 11 months. There are two main reasons for this change. First, although recruitment has been successful (approximately 600 families to date), the recruitment rate has been slower than anticipated. To be able to complete recruitment within a reasonable time frame, we need to increase the recruitment rate. Extending the potential pool of participants would be one simple way to achieve this. Second, during recruitment, a large minority of potential participants have been parents of children between the ages of 12 and 16 years. They are currently not able to take part in the study, but have expressed an interest to do so.

This change to the upper age for recruitment is the only amendment we are proposing. The revised documents include the Protocol, the text used for a simple advertisement and the information sheets for parents. The information sheets also include revised cosmetic appearance that matches the look of the information provided within the online survey. We have also taken the opportunity to update the contact information for a person not on the research team who is available to talk to parents about their potential participation in the research. Additionally, we have updated the complaints contact information on the information sheets.

No other changes have been made to the protocol, except for updating of contact information. This proposed amendment does not change the rationale/purpose for the study or any other element of the study procedure.

Any other relevant information

Applicants may indicate any specific issues relating to the amendment, on which the opinion of a reviewing body is sought.

List of enclosed documents

Notice of Amendment

IRAS Version 5.4.0

<i>Document</i>	<i>Version</i>	<i>Date</i>
Secondary caregiver study information	1.2	13/01/2017
Primary caregiver study information	1.2	13/01/2017
Project advertisement text	1.2	13/01/2017
Cerebra 1000 Families Project Protocol v1.1 6.1.17	1.1	13/01/2017

Declaration by Chief Investigator

1. *I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.*
2. *I consider that it would be reasonable for the proposed amendment to be implemented.*

This section was signed electronically by Professor Richard Hastings on 10/02/2017 00:02.

Job Title/Post: Professor
 Organisation: University of Warwick
 Email: [REDACTED]

Declaration by the sponsor's representative

I confirm the sponsor's support for this substantial amendment.

This section was signed electronically by Mrs Jane Prewett on 08/02/2017 12:12.

Job Title/Post: Head of Research Governance
 Organisation: University of Warwick
 Email: [REDACTED]

4.12: Ethics Amendment Approval Notification



Health Research Authority

West Midlands - South Birmingham Research Ethics Committee

The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

15 February 2017

Mikeda Jess
PhD Student
Centre for Educational Development, Appraisal & Research
University of Warwick
Coventry
CV4 7AL

Dear Mikeda Jess,

Study title:	The 1,000 families study: Well-being in families of children with intellectual disability
REC reference:	15/WM/0267
Amendment number:	SA 1
Amendment date:	13 February 2017
IRAS project ID:	169882

Thank you for submitting the above amendment, which was received on 13 February 2017. I can confirm that this is a valid notice of a substantial amendment and will be reviewed by the South Birmingham Sub-Committee at its next meeting.

Documents received

The documents to be reviewed are as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Copies of advertisement materials for research participants	1.2	13 February 2017
Notice of Substantial Amendment (non-CTIMP)	SA 1	13 February 2017
Participant information sheet (PIS) [Secondary Carer]	1.2	13 February 2017
Participant information sheet (PIS) [Primary Carer]	1.2	13 February 2017
Research protocol or project proposal	1.1	06 January 2017

Notification of the Committee's decision

The Committee will issue an ethical opinion on the amendment within a maximum of 35 days from the date of receipt.

R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval for the research.

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

15/WM/0267:

Please quote this number on all correspondence

Yours sincerely



Daniella Sarno
REC Assistant

Email: nrescommittee.westmidlands-southbirmingham@nhs.net

Copy to: *Professor Richard Hastings*
Ms Jane Prewett

HAYDEN, NIKITA (PGR)

From: Hayden, Nikita
Sent: 17 June 2019 14:13
To: Hayden, Nikita; Murray, Caitlin
Subject: FW: IRAS 169882. Confirmation of Amendment Assessment

From: ASHWORTH, Eleanor (HEALTH RESEARCH AUTHORITY) [<mailto:eleanor.ashworth@nhs.net>]
Sent: 07 March 2017 15:27
To: Jess, Mikeda <M.Jess@warwick.ac.uk>
Cc: Hastings, Richard <R.Hastings@warwick.ac.uk>; Sponsorship Resource <Sponsorship@warwick.ac.uk>
Subject: IRAS 169882. Confirmation of Amendment Assessment

Dear Mikeda Jess,

Further to the below, I am pleased to confirm that HRA Approval has been issued for the referenced amendment, following assessment against the HRA criteria and standards.

The sponsor should now work collaboratively with participating NHS organisations in England to implement the amendment as per the below categorisation information. This email may be provided by the sponsor to participating organisations in England to evidence that the amendment has HRA Approval.

Please contact hra.amendments@nhs.net for any queries relating to the assessment of this amendment.

Kind regards,
 Eleanor



Dr Eleanor Ashworth | HRA Assessment
Health Research Authority
 Third Floor, Barlow House, 4 Minshull Street, Manchester M1 3DZ
 E: eleanor.ashworth@nhs.net
www.hra.nhs.uk

Would you like to receive the latest updates on HRA work? Sign up [here](#)

For more information on the HRA Approval process [Click here](#)

From: nrescommittee.westmidlands-southbirmingham@nhs.net [<mailto:nrescommittee.westmidlands-southbirmingham@nhs.net>]
Sent: 15 February 2017 10:43
To: M.Jess@warwick.ac.uk
Cc: r.hastings@warwick.ac.uk; wmssponsorship@warwick.ac.uk
Subject: IRAS 169882. Confirmation of REC Validation and Categorisation of Amendment

Dear Mikeda Jess,

IRAS Project ID:	169882
REC Reference:	15/WM/0267
Short Study Title:	The 1,000 Families Study

Date complete amendment submission received:	13 February 2017
Amendment No./ Sponsor Ref:	SA 1
Amendment Date:	13 February 2017
Amendment Type:	Substantial

Thank you for submitting the above referenced amendment. I am pleased to confirm that this amendment has been submitted to the REC for ethical review. Please find attached a copy of the validation letter.

Category of Amendment

In line with the [UK Process for Handling UK Study Amendments](#) I can confirm that this amendment has been categorised as:

- **Category A** - An amendment that has implications for, or affects, ALL participating NHS organisations

You should now provide this email, together with the amended documentation, to the research management support offices **and** local research teams at your participating NHS organisations in England. If you have participating NHS organisations in Northern Ireland, Scotland and/or Wales, you should communicate directly with the relevant research teams to prepare them for implementing the amendment, as per the instructions below. You do not need to provide this email or your amended documentation to their research management support offices, as we will pass these to the relevant national coordinating functions who will do this on your behalf.

Subject to the three conditions below, you will be able to implement the amendment at your participating NHS organisations in England **35 days after you notify them of the amendment**. A template email to notify participating NHS organisations in England is provided [here](#).

- You may not implement this amendment until and unless you receive all required regulatory approvals, including REC favourable opinion, (for participating organisations in England, this includes receiving confirmation of HRA Approval for the amendment). You should provide regulatory approvals to the research management support offices and local research teams at your participating NHS organisations in England, plus to local research teams at any participating NHS organisations in Northern Ireland, Scotland or Wales*.
- You may not implement this amendment at any participating NHS organisations which inform you within the 35 day period that they require additional time to consider the amendment, until they notify you that the considerations have been satisfactorily completed.
- You may not implement this amendment at any participating NHS organisation that informs you that it is no longer able to undertake this study.

Note: you may only implement changes described in the amendment notice or letter.

If you receive required regulatory approvals (for participating organisations in England, this includes confirmation that the amendment has been granted HRA Approval) after the 35 days have passed, you may then immediately implement this amendment at all participating NHS organisations that have not requested additional review time, or are no longer able to undertake this study.

There is no need for you to receive a letter of confirmation from the participating organisation that the amendment can be implemented, as the intended date of implementation is communicated through the above process. However, you may be able to implement this amendment ahead of the 35 day deadline, if all necessary regulatory approvals are in place and the participating organisation has confirmed that the amendment may be implemented ahead of the 35 day date.

* Where the study involves NHS organisations in Northern Ireland, Scotland or Wales, the HRA will forward regulatory approvals to the relevant national coordinating function to distribute to their research management support offices.

Please do not hesitate to contact me if you require further information.

Kind regards
Daniella Sarno
REC Assistant



Health Research Authority
HRA, Ground Floor, Skipton House, 80 London Road, London, SE1 6LH
E: hra.amendments@nhs.net

www.hra.nhs.uk

The HRA is keen to know your views on the service you received – our short feedback form is available [here](#)

This message may contain confidential information. If you are not the intended recipient please inform the sender that you have received the message in error before deleting it. Please do not disclose, copy or distribute information in this e-mail or take any action in reliance on its contents: to do so is strictly prohibited and may be unlawful.

Thank you for your co-operation.

NHSmail is the secure email and directory service available for all NHS staff in England and Scotland
NHSmail is approved for exchanging patient data and other sensitive information with NHSmail and GSi recipients
NHSmail provides an email address for your career in the NHS and can be accessed anywhere
For more information and to find out how you can switch, visit www.nhsdigital.nhs.uk/nhsmail

4.13: Measure Information: Sibling Relationship Questionnaire

UNIVERSITY OF DENVER
Department of Psychology
Frontier Hall
Denver, Colorado 80208
303-871-2478

Enclosed you will find a copy of the Sibling Relationship Questionnaire (SRQ). I would be pleased to have you use it, but I do have two requests.

- 1) You may only want to use certain scales. I do not mind this kind of reduction, but I would appreciate it if the scales that are used are kept intact (i.e., not reducing the number of items to one or two or rewriting specific items). These kinds of changes make it difficult to compare results.
- 2) I would appreciate receiving information about the results of your work.

I hope you find these scales useful. This letter gives you permission to use the questionnaire. Good luck with your research!

Sincerely,

Wyndol Furman, Ph.D.
Professor

INSTRUCTIONS FOR SCORING SIBLING RELATIONSHIP QUESTIONNAIRE

Two versions of the SRQ exist. First, there is the standard 48-item version which is used when scale scores are desired. Then there is a shorter 39-item brief version which can be used if one is only interested in deriving factor scores.

Parallel versions exist for parents and children. The scale names and item compositions for the **standard version** go as follows:

Prosocial	1	17	33
Maternal Partiality	2	18	34
Nurturance of Sibling	3	19	35
Nurturance by Sibling	4	20	36
Dominance of Sibling	5	21	37
Dominance by Sibling	6	22	38
Paternal Partiality	7	23	39
Affection	8	24	40
Companionship	9	25	41
Antagonism	10	26	42
Similarity	11	27	43
Intimacy	12	28	44
Competition	13	29	45
Admiration of Sibling	14	30	46
Admiration by Sibling	15	31	47
Quarreling	16	32	48

The scale names and item compositions for the **brief version** delete items 40 – 48.

Prosocial	1	17	33
Maternal Partiality	2	18	34
Nurturance of Sibling	3	19	35
Nurturance by Sibling	4	20	36
Dominance of Sibling	5	21	37
Dominance by Sibling	6	22	38
Paternal Partiality	7	23	39
Affection	8	24	
Companionship	9	25	
Antagonism	10	26	
Similarity	11	27	
Intimacy	12	28	
Competition	13	29	
Admiration of Sibling	14	30	
Admiration by Sibling	15	31	
Quarreling	16	32	

Scale scores are derived by simply averaging the three items, except in the case of parental partiality in which case the deviations of scores of the three are averaged. In some instances an investigator may want to know the direction of parental partiality, at which point it would be appropriate to simply average the items. We have permitted subjects to miss one item on a scale and still receive the scale score in the full version. The omission of two items would result in missing data for that scale.

The derivation of factor scores is not straightforward because the structure is not simple. However, we have derived our factor scores on the basis of primary loadings. This means that the Warmth/Closeness factor consists of scale scores for intimacy, prosocial behavior, companionship, similarity, admiration by sibling, admiration of sibling, and affection. Factor scores for Relative Status/Power consist of nurturance of sibling, dominance of sibling, minus the scale scores of nurturance by sibling and dominance by sibling. Conflict scores consist of quarreling, antagonism, and competition. The Rivalry score consists of maternal and paternal partiality. In deriving factor scores, we have averaged relevant scale scores.

It is perfectly acceptable to me if you reduce the number of scales for your research purposes, but I request that you use the full number of items for each scale to ensure comparability across studies.

If you have any further questions, please feel free to contact me.

ID # _____

GROUP _____

Sibling Relationship Questionnaire - Revised (Child) 3/90

My name is _____ (completed by)

The phrase "this sibling" refers to _____ (completed about)

1. Some siblings do nice things for each other a lot, while other siblings do nice things for each other a little. How much do both you and this sibling do nice things for each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
2. Who usually gets treated better by your mother, you or this sibling?	<input type="checkbox"/> My sibling almost always gets treated better <input type="checkbox"/> My sibling often gets treated better <input type="checkbox"/> We get treated about the same <input type="checkbox"/> I often get treated better <input type="checkbox"/> I almost always get treated better
3. How much do you show this sibling how to do things he or she doesn't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
4. How much does this sibling show you how to do things you don't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
5. How much do you tell this sibling what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
6. How much does this sibling tell you what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

7. Who usually gets treated better by your father, you or this sibling?	<input type="checkbox"/> My sibling almost always gets treated better <input type="checkbox"/> My sibling often gets treated better <input type="checkbox"/> We get treated about the same <input type="checkbox"/> I often get treated better <input type="checkbox"/> I almost always get treated better
8. Some siblings care about each other a lot while other siblings don't care about each other that much. How much do you and this sibling care about each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
9. How much do you and this sibling go places and do things together?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
10. How much do you and this sibling insult and call each other names?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
11. How much do you and this sibling like the same things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
12. How much do you and this sibling tell each other everything?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
13. Some siblings try to out-do or beat each other at things a lot, while other siblings try to out-do each other a little. How much do you and this sibling try to out-do each other at things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
14. How much do you admire and respect this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

15. How much does this sibling admire and respect you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
16. How much do you and this sibling disagree and quarrel with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
17. Some siblings cooperate a lot, while other siblings cooperate a little. How much do you and this sibling cooperate with other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
18. Who gets more attention from your mother, you or this sibling?	<input type="checkbox"/> My sibling almost always gets more attention <input type="checkbox"/> My sibling often gets more attention <input type="checkbox"/> We get about the same amount of attention <input type="checkbox"/> I often get more attention <input type="checkbox"/> I almost always get more attention
19. How much do you help this sibling with things he or she can't do by him or herself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
20. How much does this sibling help you with things you can't do by yourself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
21. How much do you make this sibling do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
22. How much does this sibling make you do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

23. Who gets more attention from your father, you or this sibling?	<input type="checkbox"/> My sibling almost always gets more attention <input type="checkbox"/> My sibling often gets more attention <input type="checkbox"/> We get about the same amount of attention <input type="checkbox"/> I often get more attention <input type="checkbox"/> I almost always get more attention
24. How much do you and this sibling love each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
25. Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do you and this sibling play around and have fun with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
26. How much are you and this sibling mean to each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
27. How much do you and this sibling have in common?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
28. How much do you and this sibling share secrets and private feelings?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
29. How much do you and this sibling compete with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
30. How much do you look up to and feel proud of this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

31. How much does this sibling look up to and feel proud of you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
32. How much do you and this sibling get mad at and get in arguments with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
33. How much do both you and your sibling share with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
34. Who does your mother usually favor, you or this sibling?	<input type="checkbox"/> My sibling almost always is favored <input type="checkbox"/> My sibling is often favored <input type="checkbox"/> Neither of us is favored <input type="checkbox"/> I am often favored <input type="checkbox"/> I am almost always favored
35. How much do you teach this sibling things that he or she doesn't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
36. How much does this sibling teach you things that you don't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
37. How much do you order this sibling around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
38. How much does this sibling order you around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
39. Who does your father usually favor, you or this sibling?	<input type="checkbox"/> My sibling almost always is favored <input type="checkbox"/> My sibling is often favored <input type="checkbox"/> Neither of us is favored <input type="checkbox"/> I am often favored <input type="checkbox"/> I am almost always favored

40. How much is there a strong feeling of affection (love) between you and this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
41. Some kids spend lots of time with their siblings, while others don't spend so much. How much free time do you and this sibling spend together?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
42. How much do you and this sibling bug and pick on each other in mean ways?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
43. How much are you and this sibling alike?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
44. How much do you and this sibling tell each other things you don't want other people to know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
45. How much do you and this sibling try to do things better than each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
46. How much do you think highly of this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
47. How much does this sibling think highly of you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
48. How much do you and this sibling argue with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

ID # _____

GROUP _____

Sibling Relationship Questionnaire - Revised (Parent) 3/90

This questionnaire was completed by mother/father (circle one)

The phrase "this sibling" refers to _____

Blank lines refer to _____

1. Some siblings do nice things for each other a lot, while other siblings do nice things for each other a little. How much do both _____ and this sibling do nice things for each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
2. Who usually gets treated better by mother, _____ or this sibling?	<input type="checkbox"/> This sibling almost always gets treated better <input type="checkbox"/> This sibling often gets treated better <input type="checkbox"/> The children get treated about the same <input type="checkbox"/> _____ often gets treated better <input type="checkbox"/> _____ almost always gets treated better
3. How much does _____ show this sibling how to do things he or she doesn't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
4. How much does this sibling show _____ how to do things he or she doesn't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
5. How much does _____ tell this sibling what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
6. How much does this sibling tell _____ what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

7. Who usually gets treated better by father, _____ or this sibling?	<input type="checkbox"/> This sibling almost always gets treated better <input type="checkbox"/> This sibling often gets treated better <input type="checkbox"/> The children get treated about the same <input type="checkbox"/> _____ often gets treated better <input type="checkbox"/> _____ almost always gets treated better
8. Some siblings care about each other a lot while other siblings don't care about each other that much. How much do _____ and this sibling care about each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
9. How much do _____ and this sibling go places and do things together?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
10. How much do _____ and this sibling insult and call each other names?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
11. How much do _____ and this sibling like the same things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
12. How much do _____ and this sibling tell each other everything?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
13. Some siblings try to out-do or beat each other at things a lot, while other siblings try to out-do each other a little. How much do _____ and this sibling try to out-do each other at things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
14. How much does _____ admire and respect this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

15. How much does this sibling admire and respect _____?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
16. How much do _____ and this sibling disagree and quarrel with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
17. Some siblings cooperate a lot, while other siblings cooperate a little. How much do _____ and this sibling cooperate with other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
18. Who gets more attention from mother, _____ or this sibling?	<input type="checkbox"/> This sibling almost always gets more attention <input type="checkbox"/> This sibling often gets more attention <input type="checkbox"/> The children get about the same amount of attention <input type="checkbox"/> _____ often gets more attention <input type="checkbox"/> _____ almost always gets more attention
19. How much does _____ help this sibling with things he or she can't do by him or herself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
20. How much does this sibling help _____ with things he or she can't do by him or herself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
21. How much does _____ make this sibling do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
22. How much does this sibling make _____ do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

23. Who gets more attention from father, _____ or this sibling?	<input type="checkbox"/> This sibling almost always gets more attention <input type="checkbox"/> This sibling often gets more attention <input type="checkbox"/> The children get about the same amount of attention <input type="checkbox"/> _____ often gets more attention <input type="checkbox"/> _____ almost always gets more attention
24. How much do _____ and this sibling love each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
25. Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do _____ and this sibling play around and have fun with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
26. How much are _____ and this sibling mean to each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
27. How much do _____ and this sibling have in common?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
28. How much do _____ and this sibling share secrets and private feelings?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
29. How much do _____ and this sibling compete with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
30. How much does _____ look up to and feel proud of this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

31. How much does this sibling look up to and feel proud of _____?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
32. How much do _____ and this sibling get mad at and get in arguments with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
33. How much do both _____ and this sibling share with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
34. Who does mother usually favor, _____ or this sibling?	<input type="checkbox"/> This sibling almost always is favored <input type="checkbox"/> This sibling often is favored <input type="checkbox"/> Neither of the children is favored <input type="checkbox"/> _____ is often favored <input type="checkbox"/> _____ is almost always favored
35. How much does _____ teach this sibling things that he or she doesn't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
36. How much does this sibling teach _____ things that he or she doesn't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
37. How much does _____ order this sibling around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
38. How much does this sibling order _____ around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

39. Who does father usually favor, _____ or this sibling?	<input type="checkbox"/> This sibling almost always is favored <input type="checkbox"/> This sibling is often favored <input type="checkbox"/> Neither of the children is favored <input type="checkbox"/> _____ often is favored <input type="checkbox"/> _____ almost always is favored
40. How much is there a strong feeling of affection (love) between _____ and this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
41. Some kids spend lots of time with their siblings, while others don't spend so much. How much free time do _____ and this sibling spend together?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
42. How much do _____ and this sibling bug and pick on each other in mean ways?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
43. How much are _____ and this sibling alike?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
44. How much do _____ and this sibling tell each other things they don't want other people to know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
45. How much do _____ and this sibling try to do things better than each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
46. How much does _____ think highly of this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
47. How much does this sibling think highly of _____?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH

48. How much do _____ and this sibling argue with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> EXTREMELY MUCH
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4.14: Measure Information: Strengths and Difficulties Questionnaire

20 June 2016

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Scoring the Strengths & Difficulties Questionnaire for age 4-17 or 18+

The 25 items in the SDQ comprise 5 scales of 5 items each. It is usually easiest to score all 5 scales first before working out the total difficulties score. 'Somewhat True' is always scored as 1, but the scoring of 'Not True' and 'Certainly True' varies with the item, as shown below scale by scale. For each of the 5 scales the score can range from 0 to 10 if all items were completed. These scores can be scaled up pro-rata if at least 3 items were completed, e.g. a score of 4 based on 3 completed items can be scaled up to a score of 7 (6.67 rounded up) for 5 items.

Note that the items listed below are for 4-17-year-olds, but the scoring instructions are identical for the similarly-worded '18+' SDQ

Table 1: Scoring symptom scores on the SDQ for 4-17 year olds

	Not True	Somewhat True	Certainly True
Emotional problems scale			
ITEM 3: Often complains of headaches... (<i>I get a lot of headaches...</i>)	0	1	2
ITEM 8: Many worries... (<i>I worry a lot</i>)	0	1	2
ITEM 13: Often unhappy, downhearted... (<i>I am often unhappy...</i>)	0	1	2
ITEM 16: Nervous or clingy in new situations... (<i>I am nervous in new situations...</i>)	0	1	2
ITEM 24: Many fears, easily scared (<i>I have many fears...</i>)	0	1	2
Conduct problems Scale			
ITEM 5: Often has temper tantrums or hot tempers (<i>I get very angry</i>)	0	1	2
ITEM 7: Generally obedient... (<i>I usually do as I am told</i>)	2	1	0
ITEM 12: Often fights with other children... (<i>I fight a lot</i>)	0	1	2
ITEM 18: Often lies or cheats (<i>I am often accused of lying or cheating</i>)	0	1	2
ITEM 22: Steals from home, school or elsewhere (<i>I take things that are not mine</i>)	0	1	2
Hyperactivity scale			
ITEM 2: Restless, overactive... (<i>I am restless...</i>)	0	1	2
ITEM 10: Constantly fidgeting or squirming (<i>I am constantly fidgeting...</i>)	0	1	2
ITEM 15: Easily distracted, concentration wanders (<i>I am easily distracted</i>)	0	1	2
ITEM 21: Thinks things out before acting (<i>I think before I do things</i>)	2	1	0
ITEM 25: Sees tasks through to the end... (<i>I finish the work I am doing</i>)	2	1	0
Peer problems scale			
ITEM 6: Rather solitary, tends to play alone (<i>I am usually on my own</i>)	0	1	2
ITEM 11: Has at least one good friend (<i>I have one good friend or more</i>)	2	1	0
ITEM 14: Generally liked by other children (<i>Other people my age generally like me</i>)	2	1	0
ITEM 19: Picked on or bullied by other children... (<i>Other children or young people pick on me</i>)	0	1	2
ITEM 23: Gets on better with adults than with other children (<i>I get on better with adults than with people my age</i>)	0	1	2
Prosocial scale			
ITEM 1: Considerate of other people's feelings (<i>I try to be nice to other people</i>)	0	1	2
ITEM 4: Shares readily with other children... (<i>I usually share with others</i>)	0	1	2
ITEM 9: Helpful if someone is hurt... (<i>I am helpful if someone is hurt...</i>)	0	1	2
ITEM 17: Kind to younger children (<i>I am kind to younger children</i>)	0	1	2
ITEM 20: Often volunteers to help others... (<i>I often volunteer to help others</i>)	0	1	2

20 June 2016

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Total difficulties score: This is generated by summing scores from all the scales except the prosocial scale. The resultant score ranges from 0 to 40, and is counted as missing if one of the 4 component scores is missing.

'Externalising' and 'internalising' scores: The externalising score ranges from 0 to 20 and is the sum of the conduct and hyperactivity scales. The internalising score ranges from 0 to 20 and is the sum of the emotional and peer problems scales. Using these two amalgamated scales may be preferable to using the four separate scales in community samples, whereas using the four separate scales may add more value in high-risk samples (see Goodman & Goodman, 2009 *Strengths and difficulties questionnaire as a dimensional measure of child mental health. J Am Acad Child Adolesc Psychiatry* 48(4), 400-403).

Generating impact scores

When using a version of the SDQ that includes an 'impact supplement', the items on overall distress and impairment can be summed to generate an impact score that ranges from 0 to 10 for parent- and self-report, and from 0 to 6 for teacher-report.

Table 2: Scoring the SDQ impact supplement

	Not at all	Only a little	A medium amount	A great deal
<u>Parent report:</u>				
Difficulties upset or distress child	0	0	1	2
Interfere with HOME LIFE	0	0	1	2
Interfere with FRIENDSHIPS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
Interfere with LEISURE ACTIVITIES	0	0	1	2
<u>Teacher report:</u>				
Difficulties upset or distress child	0	0	1	2
Interfere with PEER RELATIONS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
<u>Self-report report:</u>				
Difficulties upset or distress child	0	0	1	2
Interfere with HOME LIFE	0	0	1	2
Interfere with FRIENDSHIPS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
Interfere with LEISURE ACTIVITIES	0	0	1	2

Responses to the questions on chronicity and burden to others are not included in the impact score. When respondents have answered 'no' to the first question on the impact supplement (i.e. when they do not perceive themselves as having any emotional or behavioural difficulties), they are not asked to complete the questions on resultant distress or impairment; the impact score is automatically scored zero in these circumstances.

Cut-points for SDQ scores for age 4-17: original 3-band solution & newer 4-band solution

Although SDQ scores can be used as continuous variables, it is sometimes convenient to categorise scores. The initial bandings presented for the SDQ scores were 'normal', 'borderline' and 'abnormal'. These bandings were defined based on a population-based UK survey, attempting to choose cutpoints such that 80% of children scored 'normal', 10% 'borderline' and 10% 'abnormal'.

More recently a four-fold classification has been created based on an even larger UK community sample. This four-fold classification differs from the original in that it (1) divided the top 'abnormal' category into two groups, each containing around 5% of the population, (2) renamed the four categories (80% 'close to average', 10% 'slightly raised', 5% 'high' and 5% 'very high' for all scales except prosocial, which is 80% 'close to average', 10% 'slightly lowered', 5% 'low' and 5% 'very low'), and (3) changed the cut-points for some scales, to better reflect the proportion of children in each category in the larger dataset.

Note that these cut points have not been validated for use with the 18+ SDQ, so we suggest that it is safest to use continuous scores rather than categories for this measure

Table 3: Categorising SDQ scores for 4-17 year olds (not validated for 18+)

	Original 3-band categorisation			Newer 4-band categorisation			
	Normal	Borderline	Abnormal	Close to average	Slightly raised (/slightly lowered)	High (/Low)	Very high (very low)
<u>Parent completed SDQ</u>							
Total difficulties score	0-13	14-16	17-40	0-13	14-16	17-19	20-40
Emotional problems score	0-3	4	5-10	0-3	4	5-6	7-10
Conduct problems score	0-2	3	4-10	0-2	3	4-5	6-10
Hyperactivity score	0-5	6	7-10	0-5	6-7	8	9-10
Peer problems score	0-2	3	4-10	0-2	3	4	5-10
Prosocial score	6-10	5	0-4	8-10	7	6	0-5
Impact score	0	1	2-10	0	1	2	3-10
<u>Teacher completed SDQ</u>							
Total difficulties score	0-11	12-15	16-40	0-11	12-15	16-18	19-40
Emotional problems score	0-4	5	6-10	0-3	4	5	6-10
Conduct problems score	0-2	3	4-10	0-2	3	4	5-10
Hyperactivity score	0-5	6	7-10	0-5	6-7	8	9-10
Peer problems score	0-3	4	5-10	0-2	3-4	5	6-10
Prosocial score	6-10	5	0-4	6-10	5	4	0-3
Impact score	0	1	2-6	0	1	2	3-6
<u>Self-completed SDQ</u>							
Total difficulties score	0-15	16-19	20-40	0-14	15-17	18-19	20-40
Emotional problems score	0-5	6	7-10	0-4	5	6	7-10
Conduct problems score	0-3	4	5-10	0-3	4	5	6-10
Hyperactivity score	0-5	6	7-10	0-5	6	7	8-10
Peer problems score	0-3	4-5	6-10	0-2	3	4	5-10
Prosocial score	6-10	5	0-4	7-10	6	5	0-4
Impact score	0	1	2-10	0	1	2	3-10

Note that both these systems only provide a rough-and-ready way of screening for disorders; combining information from SDQ symptom and impact scores from multiple informants is better, but still far from perfect.

