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Maternal Positivity in Mothers Raising Children with ID

By

Mikeda Jess

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List of Abbreviations

AIC	Akaike Information Criterion
ASBAH	Association for Spina Bifida and Hydrocephalus
ASD	Autism Spectrum Disorder
BPI-01	The Behavior Problems Inventory
CFA	Confirmatory Factor Analysis
CFI	Confidence Fit Index
CMIN/DF	the ratio of chi-square to degrees of freedom
CPHP	Chronic Physical Health Problem
CPRS	The Child-Parent Relationship Scale
DBC-P	The Developmental Behaviour Checklist- Parent
DD	Developmental Disabilities
DS	Down's syndrome
ECVI	Expected Cross-Validation Index
FIQ	Family Index Questionnaire
HADS	Hospital Anxiety and Depression Scale
ID	Intellectual Disability
K6	Kesslar 6

KIPP-PC/ PCS	Kansas Inventory of Parental Perceptions Positive Contributions/ Positive Contributions Scale
MGCFA	Multigroup Confirmatory Factor Analyses
MP	Maternal Positivity
NCBRF	The Nisonger Child Behaviour Rating Form
NHS	National Health Service
NIPT	Non-Invasive Prenatal Testing
PANAS	Positive and Negative Affect Schedule
PGS	Positive Gains Scale
QRS-F	Questionnaire on Resources and Stress (Friedrich, short form)
RMSEA	Root Mean Square Error of Approximation
SDQ	Strengths and Difficulties Questionnaire
SEM	Structural Equation Modelling
SES	Socio-Economic Status
SWLS	Satisfaction with Life Scale
TBPS	Total Behaviour Problem Score
TD	Typically Developing
VABS II	The Vineland Adaptive Behaviour Scale II- Survey form

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Thesis Abstract

Background: Raising a child with Intellectual Disability (ID) is a unique experience in which families are faced with many challenges. However, despite this, mothers raising children with ID do report having positive perceptions of their child and perceive their child as having a positive impact on themselves and wider family members. This thesis explored the positivity of mothers raising children with an Intellectual Disability (ID).

Method: Chapter 2 incorporated the use of Structural Equation Modelling (SEM) to create a latent construct of maternal positivity using single indicators of positivity. Chapter 3 investigated the function of maternal positivity within the context of resilience. Chapter 4 explored the phenomenon of the Down's syndrome (DS) advantage and Chapter 5 explored whether the measure of positive gains operates similarly across parents of children with ID, parents of children with a physical health condition, and parents of typically developing children. Each of the studies are designed and written as empirical papers to be published. Chapter 1 describes the background to the thesis and chapter 6 presents an overall discussion of the thesis.

Results: Chapter 2 found that the latent construct of positivity had significant relationships with both child and maternal outcomes. Chapter 3 evidenced that maternal positivity could be described as a resilience variable that had a largely compensatory function cross-sectionally. Chapter 4 found that the Down's syndrome advantage was only evident for maternal positive gains when covariates were accounted for. Finally, chapter 5 evidenced that the Positive Gains Scale (PGS) means were not equivalent across the three study groups indicating that valid mean comparisons could not be made.

Conclusion: Maternal positivity exists for mothers raising children with ID alongside elevated levels of maternal stress and psychological distress and appears to function as a resilience variable. The

Down's syndrome advantage was evident for maternal positive gains and chapter 5 demonstrated the importance of ensuring measurement invariance when making mean comparisons between groups. A more in-depth overview of the thesis conclusions are discussed in chapter 6.

Chapter 1: Introduction and overview

Definition of Intellectual Disability (ID)

Intellectual Disability (ID) is defined by the American Association on Intellectual and developmental disabilities as, "...a disability characterised by significant limitations in both intellectual functioning and in adaptive behaviour, which covers many everyday social and practical skills. This disability originates before the age of 18". Adaptive behaviours covers a range of everyday and practical skills. These include social skills, the ability to react appropriately to interpersonal social cues and daily living skills such as personal self-care and occupational skills. There are multiple reasons why a child would have ID. A diagnosis of ID is sometimes caused by abnormal genes inherited from their parents or errors when genes combine. Examples of genetic conditions include Down's syndrome and Fragile X syndrome. ID can also result from problems during pregnancy and problems during birth. Children with ID will generally experience more challenges than a typically developing child.

Challenges Faced by Families

Research has consistently demonstrated that parents raising a child with Intellectual Disability are likely to experience elevated levels of anxiety, parenting stress and depression compared to parents of typically developing children. In a longitudinal study Baxter and colleagues (2000) found that the stress parents attributed to family members with disabilities was around twice of that attributed to the sibling without a disability.

Raising a child with Intellectual Disability brings about a unique set of challenges and these quite often have an impact on all family members. Although caregiving is expected when raising a child this role has a unique significance when a child has functional limitations and long-term dependence. One main challenge for families is to manage the disabled child's needs effectively whilst still observing the requirements of everyday living. In some cases, the provision of such demanding care can prove detrimental to both the physical health and the psychological well-being of parents of disabled children which can

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have an impact on family income, family functioning, and sibling adjustment (King, King, Rosenbaum & Goffin;1999).

The poor mental health reported by parents of children with ID may place parents at an increased risk of marital disruption, family dysfunction and mental health problems (McConnell & Savage, 2015). There is extensive literature focused on the negative adjustment of both siblings of children with ID and their parents (Dyke, Mulroy & Leonard, 2009; Meppelder, Hodes, Kef & Schuengel, 2015; Olsson & Hwang, 2001; Patton, Ware, McPherson, Emerson & Lennox, 2018; Shivers, McGregor, & Hough, 2017). Findings from these studies suggest that multiple factors such as parental cognitions (Lloyd & Hastings, 2009), socio-economic factors (Emerson, 2003), severity of child diagnosis (Hastings & Johnson, 2001), child behaviour problems (Beck, Hastings, Daley & Stevenson, 2004) and social support (Davis & Gavida-Payne, 2009), all have an effect on the well-being of the whole family.

Disabled children and their families have reported that they find it difficult to access leisure facilities and transport that can adequately accommodate their needs (Beart, Hawkins, Kroese, Smithson & Tolosa; 2001; Shikako-Thomas, Majnemer, Law & Lach; 2008). Consequently, this limits participation in recreational activities as a family unit which would arguably have a negative impact on family cohesion.

Furthermore, impaired social interactions, challenging behaviours and low cognitive functioning are all common characteristics amongst children with ID and these characteristics often make them 'stand out'. Consequently, children with disabilities frequently experience stigma which extends to their family members. Many families raising children with disabilities have reported experiencing both enacted and felt stigma. Enacted stigma refers to instances where overt rejection or discrimination is experienced by stigmatised individuals and felt stigma refers to the feelings of shame and or the feeling of rejection (Gray, 2002). In

a qualitative study, Gray (2002) found that most parents experienced stigma and that felt stigma was most commonly experienced in public situations such as social outings. Importantly, parents reported frequently not being invited to social occasions which had a negative effect on the whole family. In turn, many families restricted their social life to avoid the negative reactions of others, a decision that families raising a child without disabilities would not have to consider.

Mothers and Stressors

It is widely acknowledged that poor parental mental health is more prevalent in mothers than fathers (Dabrowska & Pisula, 2010; Norlin & Broberg, 2013; Olsson & Hwang, 2001). One reason for this consistent finding could be that in most cases mothers take on the majority of the childcare and practical responsibility pertaining to their child with ID which are often far greater than those placed on mothers of children without disabilities (Shearn & Todd, 2000). Therefore, any adverse or beneficial experiences associated with raising a child would often impact the mother more so than other family members. The differential outcomes may also reflect the greater amount of time mothers spend with their child compared to fathers. In addition, mothers may have had to give up their jobs in order to take care of their child and consequently feel unable to pursue their own interests (Potterfield, 2002; Shearn & Todd, 2000).

Child behaviour problems are often predictive of later adverse maternal outcomes even from a young child-age (Baker et al, 2003). Studies of mothers raising children with ID clearly demonstrate the link between elevated levels of maternal psychopathology and child characteristics. This is also evident in mothers of typically developing children. For example, in an early study, Donenberg and Baker (1993) compared the impact on parents of young typically developing (TD) children with behaviour problems and children with ASD, which is associated with increased rates of behaviour problems. Parents of TD children with behaviour

problems reported comparable levels of stress and negative impact to those reported by parents of children with ASD. Similar findings were evident in a longitudinal study of mothers of children that were either typically developing or that had ID. Neece and Baker (2008) found that child behaviour problems at age six were a significant predictor of maternal parenting stress two years later irrespective of child diagnosis. These findings demonstrate that relationships between child factors and maternal mental health are not specific to parents of children with ID. Importantly, it is clear from the research outlined above that the mental health of parents, irrespective of child diagnosis, is vulnerable to the impact of their child's adverse characteristics. However, what separates mothers of children with ID from mothers of typically developing children is that the former often have multiple child-related challenges to manage in addition to numerous behaviour problems therefore arguably, the adverse risk to mental health is greater.

Child-related factors have been found to be significant predictors of maternal well-being many years later. Grein and Glidden (2015) recently published a study which explored the well-being outcomes of mothers of children with ID over a twenty-year period. The researchers found that child cognitive impairment, an indicator of disability severity, was a significant predictor of maternal depression twenty years later.

In addition to child-related factors, several studies have reported that family cohesion, household income and support are also associated with maternal well-being. Hassall and colleagues (2005) examined the effects of support on parenting stress in mothers of children with ID. The researchers found a negative association between family support and parenting stress which indicates that mothers with higher levels of family support experience lower levels of parenting stress. These findings are consistent with Johnston and colleague's (2003) previous research on the relationship between family cohesion and maternal psychological

well-being thus indicating that support systems are important not only for the family as a whole but also specifically for mothers.

Research has also indicated that mothers raising children with ID are adversely affected by socioeconomic factors. Emerson and colleagues (2006) proposed that research examining socio-economic factors have overlooked their importance within families that include members with ID. The researchers found that mothers of children with ID reported lower levels of happiness than mothers of children without disabilities however when socioeconomic factors were controlled for, group differences no longer remained, and it was found that socioeconomic factors accounted for 50% of the increased risk for poorer maternal self-esteem and self-efficacy. In a later similar study, Olsson and Hwang (2008) found that mothers of children with ID had lower levels of well-being than mothers of children without disabilities however like the previous study, differences in economic hardship were the biggest predictor of maternal well-being. Therefore, these findings indicate that lower maternal well being cannot simply be accounted for by their child's behaviour and further suggest that other factors need to be considered.

However, there is variability in maternal outcomes and importantly, it has been reported that some mothers thrive in the face of difficulties related to their child. In some cases, although mothers have reported greater stress, depression and anxiety than fathers, they have also reported that they perceive their child more positively than fathers (Hastings, Beck & Hill, 2005). It has been suggested that positive perceptions of the child may protect maternal mental health against adverse experiences related to child characteristics (e.g. challenging behaviours), likely moderating the relationship between child-related predictors and maternal outcomes.

What is Positivity?

'Positivity' in the general domain pertains to one's experience of positive emotions such as joy, hope and happiness (Fredrickson, 2004). Positivity, by contrast, has not received the same level of empirical attention as has negativity or negative outcomes. However, within the last 30 years this has started to change and increasingly, focus has shifted to positivity and how it functions. Traditionally positive and negative emotions were viewed to be on an interrelated spectrum of emotions along a single continuum on opposite ends from high positive affect to high negative affect, however they were later recognised as two separate dimensions.

There have been increasing efforts to understand positivity and how it functions in relation to adversity. Watson and colleagues (1988) demonstrated the relationship between positive affect and poor metal health. Whilst negative affect was found to be significantly associated with symptoms of both depression and anxiety, positive affect was only associated with symptoms of depression. These findings suggest that positive affect and negative affect operate independently and that they are distinctly sperate constructs that have different relationships with different diagnoses of mental health. The findings also indicate that positivity could play a significant role in interventions aimed at reducing depressive symptomology.

In more recent times, empirical evidence supporting the view that increased positivity is predictive of improved well-being and other favourable outcomes, have started to emerge.

Evidence has repeatedly demonstrated that stress is associated with a plethora of negative outcomes such as depression, anxiety and even physical symptoms (Zautra, 2005). However, more recently focus has started to shift to positive aspects of the stress process.

Increasingly, researchers are starting to explore the mechanisms surrounding the observed positive outcomes that sometimes occur in stressful situations.

A popular framework for investigating stress is the stress and coping theory of Lazarus and Folkman (1984) which focuses on coping processes that help manage or reduce aversive states. Folkman (1997) drew on her earlier work, in which she proposed that positive emotions have important adaptive functions during stress (Lazarus, Kanner and Folkman, 1980), to develop a modified version of the coping model. In a longitudinal study, Folkman (1997) monitored gay men who were primary carers for their partner with AIDS for up to five years. Caregivers reported higher levels of depression than the general population. However, throughout the study, except for the immediate weeks preceding and following their partner's death. Caregivers also reported experiencing a positive mood at similar frequencies to their negative mood. Given the evidence that positive emotions can occur in the most stressful of situations, Folkman (1997) proposed that positive emotions, or positive reappraisal, experienced during long periods of sustained stress serve as an effective coping mechanism.

Positive reappraisal is a cognitive process through which people focus on the good of what is happening or happened previously. Through the process of positive reappraisal, the meaning of the situation has changed which allows a person to experience positive emotions and well-being (Folkman & Moskowitz, 2000). Folkman's longitudinal study found that positive reappraisal was consistently associated with positive emotions during both caregiving and after the death of their partner. Thus, Folkman's revised stress and coping theory posits that the positive emotions that co-occur with negative emotions during intensely stressful situations have an important function in the stress process in that they support effective coping processes. Folkman (1997) proposed in part that the negative psychological state associated with significant and enduring stress could motivate people to search for and create *positive* psychological states to gain relief from the distress. Furthermore, the positive

psychological states are hypothesised to help sustain problem and emotion focused coping strategies to deal with the stressful condition.

If applying Folkman's theory to mothers of children with ID, positive reappraisal processes may help mothers to redefine and focus on the positive meaning attributed to the experience of raising their child with ID which may consequently support effective coping strategies.

Resilience theory attempts to address why some people can achieve and sustain health and well-being when faced with adversity. Resilience itself can be described as being able to successfully adapt to a maladaptive situation and has been defined as "…the individual variations in response to risk. Some people succumb to stress and adversity whereas others overcome life hazards" (Rutter, 1987, p. 317). Such definitions of resilience are drawn from a risk/ stress resilience framework: for resilience to be demonstrated, a stressor must be experienced. Resilience has been conceptualised as having two main functions: a compensatory function (having a direct main effect, reducing negative outcomes directly) and a protective function (reducing negative outcomes in the context of exposure to risk – a moderating effect). Therefore, for resilience to be evident, positive adaptation must be present despite a risk (Luthar et al. 2000; Tusaie & Dyer, 2004). Thus, these conceptualisations of how resilience functions lead to varying predictions of family outcomes in family disability research.

In line with resilience theorists, families raising a child with ID will demonstrate resilience when despite being confronted with multiple stressors, such as child behaviour problems, they will still report positive outcomes pertaining to themselves and/ or their child with ID. Indeed, families raising children with disabilities do report varying outcomes, with some reporting more positively than others. This perhaps indicates that families which have

more favourable outcomes than other families score higher in variables that could be conceptualised as having a resilience function (e.g. family support). External (e.g.; support agencies) child (e.g. prosocial skills) and family variables (e.g. family cohesion) that positively correlate with positive parental well-being outcomes could potentially have a resilience function. However, these variables would need to be explicitly investigated for their putative function before any firm conclusions could be made. Therefore, furthering our understanding about which variables have either a protective or compensatory function on maternal mental health could provide valuable information pertaining to established risk variables/ stressors that are not associated with negative outcomes.

In a study of stress and coping, Gloria and Steinhardt (2014) found significantly negative associations between positive emotions and stress in addition to positive emotions and depressive symptoms which indicates that those that experience higher frequencies of positive emotions will have better mental health outcomes than those who don't. These findings further indicate that positive emotions could potentially be conceptualised as a resilience variable, having a compensatory function which could potentially be utilised to improve psychopathology. This would require further exploration however the findings do suggest that the positive emotions that may emerge during periods of sustained stress (Stress and coping theory: Folkman; 1997) could function as a resilience variable.

Although the benefits of positivity within the general population have become clearer in the last thirty years, more empirical research is needed within other populations in order to fully understand positivity and to potentially guide interventions that may improve functioning and psychological well-being. Research into positivity is grossly underrepresented in families raising children with Intellectual Disability (ID). However, the existing research suggests that positivity does exist within these families although there is much debate on how it is defined, the factors that may impact it and how it functions. Thus,

in order to address outstanding questions within the family and disability field, the focus of this thesis is on positivity in mothers raising children with Intellectual Disability (ID).

Definition of Maternal Positivity and Existing Measures within Disability Research

Historically, raising a child with a disability was largely viewed as a negative experience with predominantly negative outcomes for both the child and their parents. Researchers primarily adopted negative assumptions that were reflected in their research questions and hypotheses. Although 'positivity' makes reference to positive emotions, quite often within disability research a positive outcome would be evidenced by the absence of a negative construct (e.g. depression) or comparatively lower levels of a negative construct in a comparison design study. However, this approach does not allow the researcher to measure positivity directly.

In an earlier paper, Hastings and Taunt (2002) highlighted the lack of research that explicitly addressed positivity within the disability field. However, to be able to do this there must be a consensus of what maternal positivity looks like and how to measure it. To date, positivity has been described in various ways and therefore there is no one instrument that has been implemented which exclusively measures 'maternal positivity'. Instead researchers have used a variety of instruments to identify and measure positivity.

One approach has been to adopt the use of instruments that measure general positivity to identify the construct; that is positivity that is not specific to either parenting or to disability. The Positive and Negative Affect Schedule (PANAS) measures positivity independently of negativity and is broad in that it can be utilised across different sample groups, e.g. disabled and non-disabled. Positivity in mothers has been measured with success by utilising the positive affect scale taken from the PANAS. Satisfaction with life has also been used as in indicator of maternal positivity in disability research. This is measured using another general positivity measure, the Satisfaction with Life Scale (SWLS) created by

Diener (1985). Although these measures are helpful, their insight into positivity specific to mothers is limited. Neither instrument was designed exclusively for parents therefore they are liable to overlook key elements of positivity that may be specific to parenting. Although a mother may score highly on the SWLS, indicating high life satisfaction, it should not be assumed that this is specific to their role as a parent or indeed that they are highly positive about parenting. Other general concepts of positivity such as hope and optimism have been adopted within research in an effort to identify positivity in mothers. However, they are also limited in their ability to fully quantify positivity specific to parenting.

A number of researchers have adopted concepts specific to parenting to define maternal positivity. To feel highly efficacious in one's parenting role is a popular indicator of maternal positivity. In addition to this, a close parent-child relationship has also been adopted as an indicator of maternal positivity. The benefit of this approach is that it explicitly taps into areas of positivity that are specific to parenting. As the body of positivity research grows within the disability field, various concepts have been adopted to define maternal positivity. Arguably concepts that are specific to positivity and raising a child with a disability would be an appropriate definition of 'maternal positivity'. Although research into specific areas of maternal life offer some insight, positivity often taps onto multiple domains of a mother's life. Therefore, to gain a robust understanding of maternal positivity, it is important to consider all concepts of maternal positivity to identify what positivity looks like for mothers raising children with ID.

Previous Research on Maternal Positivity

It was identified early on that mothers raising a child with intellectual disability reported higher levels of stress and increased anxiety compared to parents raising typically developing children. Qualitative studies highlighted that in addition to the challenges involved, parents were keen to talk about the positive feelings they had about their child with a disability

(Stainton & Besser, 1998). Now with the advent of instruments that measure positive constructs, a quantitative approach to research could be explored. Importantly, maternal positivity could be measured and compared between groups.

Hastings and Taunt (2002) identified that within families raising a child with ID, positivity does exist alongside some of the negative experiences, however this was not reflected in much of the family disability literature. Conversely, they proposed a working model for the future study of positivity in families of children with ID based on the hypothesis that positive perceptions function in a way that help families adapt with the experience of raising a child with ID (Hastings & Taunt, 2002). However, to be able to understand positivity in greater detail and its function, researchers must first define what positivity is and then be able to explicitly measure positivity.

To date, there are various ways in which positivity has been defined and quantified in ID research. There are, however, a small number of instruments which have been designed to identify and measure positivity in families raising children with disabilities. The Kansas Inventory of Parental Perceptions Positive Contributions scale (KIPP-PC: Behr, Murphy & Summers, 1992) is a questionnaire that consists of 50 items which measures the positive impact that a child with a disability has on the parent and the family as a whole. The Family Impact Questionnaire (FIQ: Donenburg & Baker, 1993) also consists of 50 items but measures the positive and negative impact of a disabled child however it includes six subscales, one of which being the 'Positive Impact on Parenting' subscale that pertains to the positive impact the disabled child has on the parent compared with other children. Subsequent to the Hastings and Taunt (2002) review, the Positive Gains Scale (PGS: Pit-ten Cate, 2003) was introduced as a short concise instrument designed to specifically measure the perceived benefits of raising a child with a disability including growing as a person and becoming closer as a family.

The introduction of these instruments has enabled researchers quantifiably define maternal positivity and measure it. Subsequently studies are able to evidence that although parents of children with ID experience high levels of distress, often, these parents also perceive their child as having a positive impact on themselves and their family which appear to be separate to the degree of negativity experienced. For example, one recent study (Griffith et al, 2011) explored parental psychological distress (stress, anxiety and depression) and positive gains in three ID diagnostic groups (Angelman syndrome, Cornelia de Lange syndrome & Cri du Chat syndrome). The researchers identified differential levels of negative maternal outcomes reported between groups, however utilising the PGS it was found that the positive impact of the child on their mother and family did not differ significantly between groups suggesting that positive gains is not associated with negative maternal outcomes. Similar findings were reported by Vilaseca and colleagues (2014) who found no significant associations between maternal scores of positive perceptions of their child with ID and maternal depression and anxiety. Together, these findings are consistent with the earlier discussed general theories of positivity, in that positive and negative affect are separate constructs. Yet, the latter study indicates that positivity does not support better maternal psychological outcomes given that positivity had no relationship with maternal mental health. However, there are some existing research that contradicts this.

Kayfitz and colleagues (2010) identified significantly negative association between parenting stress and positive contribution scores for mothers of children with ASD. Hastings and colleagues (2005a) also found that mothers' positive perceptions of their child with ID were negatively associated with their parenting stress and depression but not anxiety indicating somewhat of a compensatory function. Thus, the relationship between positivity and maternal psychopathology is unclear and there is a need for a coherent overview of

associations between maternal positivity and poor maternal mental health such as parenting stress and depression.

A number of researchers within the developmental disability field have explored the relationships between maternal positivity and child behaviour. For example, MacMullin, Tint and Weiss (2011) explored positive gain in mothers of children with ASD and found no association between child behaviour problems and positive gains, indicating that there is no relationship between positivity and adverse child behaviours. However, there are existing studies which suggest otherwise. In an early study focussed on mothers of children with Down's syndrome (DS) child behaviour problems were found to have a significantly negative association with maternal satisfaction with life (Sloper et al, 1991).

It has become evident that the extent of child behaviour problems varies with diagnostic syndrome (Abbeduto et al, 2004; Blacher and McIntyre, 2006; Eisenhower, Baker & Blacher, 2005) and children with Down's syndrome (DS) are frequently reported to exhibit comparatively less behaviour problems and more prosocial behaviours. A Down's syndrome advantage refers to the view that individuals with DS and their families have better outcomes than those with other Intellectual Disabilities. Several studies have demonstrated this (Blacher et al, 2013; Hodapp et al, 2003; Pisula, 2007; Ricci & Hodapp, 2003) however few have explored positive maternal outcomes within the context of the DS advantage.

Griffith and colleagues (2010) employed a comparison design study and found that compared to children with other ID diagnoses, children with DS were reported to have the lowest levels of behaviour problems and their mothers reported the highest mean scores of positive perceptions and positive affect. The implication here is that child behaviour problems have a negative impact on positive maternal outcomes however it is important to note that this was not explicitly tested for in the study. The Down's syndrome advantage

specific to maternal stress has been partially explained by higher levels of adaptive behaviours and lower levels of problem behaviours however there is no clear consensus on whether this would be the case for positive maternal outcomes which calls for a robust overview and further study.

Finally, in some cases positivity has been associated with alleviating maternal mental health and child behaviour problems (Blacher & Baker, 2007). The aforementioned Hastings and Taunt (2002) review raised questions regarding the function of positivity and attempts to address this have been reflected in more recent studies. Unfortunately, findings have been contradictory. Whilst some studies have found that elements of maternal positivity do function in a way that reduces maternal psychopathology and protects maternal mental health from child-related risk factors, some studies have found the contrary.

Aims of the thesis

There are questions that remain unanswered within family disability literature which if addressed will advance wider knowledge surrounding maternal positivity and could potentially have important implications for clinical practice. The overarching aim of this thesis was to investigate the construct and measurement of positivity in mothers raising children with ID. In particular, I was interested in testing whether maternal positivity is a single over-arching construct (chapter 2). Further, I aimed to explore how the construct of positivity operates within a resilience framework (chapter 3), and whether certain aspects of this construct (for example positive gains) show variation depending on the aetioloy of intellectual disability (i.e., testing the Down syndrome advantage; chapter 3). Finally, I aimed to explore whether the measure of one aspect of maternal positivity (positive gains) operates in a similar way across different disability groups (Chapter 5). Below I provide further information on these chapters.

The aim of chapter 2 was to investigate whether maternal positivity can be described as one latent variable using multiple observed indicators of positivity. The secondary aim of this study was to identify potential relationships between maternal positivity and child/ maternal outcomes. Significant relationships between a latent construct of maternal positivity and maternal outcomes would indicate that maternal positivity may function as a resilience variable which has important implications for practice. The aim of chapter 3 was to investigate whether maternal positivity can be utilised to support the mental health of mothers raising children with disabilities within a resilience framework both cross-sectionally and longitudinally. There is much debate surrounding the Down's syndrome advantage. Whilst there is the argument that the advantage is solely attributable to a diagnosis of DS, those opposing suggest that the DS advantage is caused by external factors including maternal characteristics and socioeconomic factors. Furthermore, little is known about whether the DS advantage can be measured for positive maternal outcomes such as positive perceptions of their child. Therefore, the aim of chapter 4 was to investigate whether the DS advantage remains present for positive maternal outcomes when external factors have been accounted for. As proposed in the Hastings and Taunt (2002) review, group comparison studies are required to identify potential group differences in positivity. In order to effectively make those comparisons, it is essential that instruments are measuring the same construct of positivity within each group otherwise reported findings will not be valid. Therefore, the aim of chapter 5 was to investigate whether an instrument designed to measure positivity in parents raising a child with a disability worked equivalently for mothers of children belonging to two distinct disability groups and mothers of typically developing children.

Structure of This Thesis

Chapter 2 is the first study in this thesis. Although the term 'maternal positivity' is used within disability literature, there is yet to be a consensus on what maternal positivity actually

looks like. There were two clear aims of this study, the first was to create a latent construct of maternal positivity. Here, secondary data were used to create a latent construct by utilising multiple single indicators of positivity that were either general, disability- specific or parenting- specific. The second aim was to explore potential associations between the construct of maternal positivity and child and maternal outcomes. If a construct of maternal positivity could be created by indicators that tap into multiple areas of maternal life this would suggest that single indicators are not broad enough to fully represent maternal positivity in its entirety. Furthermore, if associations were identified between a

construct of maternal positivity and poor maternal mental health and child behaviours such findings would raise questions about implications for intervention for mothers raising children with ID.

Chapter 3 presents the second study of this thesis and focuses on the function of the construct of maternal positivity developed in the previous study within the context of resilience theories. Resilience has been described as the ability to withstand stress and remain competent even when exposed to stressful life events. Resilience variables have two distinct functions, compensatory and protective. Protective variables have a moderating effect and act as a buffer against a stressor or a 'risk' variable whereas a compensatory variable can be described as having a main effect, operating counteractively and improving levels of competence in the face of a stressor. Therefore if maternal positivity had a protective function then it would ameliorate the impact of the risk variable (in this case, child behaviour problems) on maternal psychological distress. Whereas if maternal positivity had a compensatory function then it would have a direct main effect and reduce levels of maternal psychological distress. The aim of this study was to investigate how maternal positivity functioned cross-sectionally and longitudinally.

Chapter 4 examines the Down's syndrome advantage which suggests that parents raising children with Down's syndrome experience better well-being outcomes, mostly lower levels of psychological distress, than parents raising children with other intellectual disabilities. There has been much debate within ID literature. Whilst it has been argued that parents of children with Down's syndrome have better outcomes due to their child's diagnoses, increasingly research has begun to explore the role of external factors such as socioeconomic status and child behaviours as potential confounding variables.

Furthermore, ID literature is lacking as to whether the Down's syndrome advantage is present in positive maternal outcomes. Using data from the 1000 families study (see Appendix I), the aim of this study was to determine whether the Down's syndrome advantage was present in positive and negative maternal outcomes once child and maternal factors were controlled. It was predicted that any evidence of a Down's syndrome advantage in this study would be due to external factors.

Chapter 5 is the final empirical study. As was proposed in the Hastings and Taunt (2002) review, comparison studies are required to identify potential group differences in positivity. In order to effectively make those comparisons it is essential that the construct being measured has the same meaning for each group otherwise comparisons are not valid. This chapter takes a closer look at the psychometric properties of the Positive Gains Scale, an instrument designed to measure the perceived positive impact a disabled child has on their caregiver and their family. Using data from the 1000 families study (see Appendix I), and secondary data on mothers of children with ASD, mothers of TD children and mothers of children with a chronic physical health problem (CPHP), the aim of this study was to determine whether the PGS operated equivalently for all groups of mothers.

All research questions were approached quantitatively and an overview of data sources can be found in Appendix XIX.

Chapter 6 concludes this thesis and discusses the studies' contribution to knowledge. It summarises the findings and methodological limitations, in addition to outlining future research and implications for clinical practice.

Orientation to Chapter 2

This study was conducted to investigate the viability of producing a latent construct of maternal positivity using five single indicators of positivity. Much of the research around positivity incorporates varying definitions of positivity and within family and disability literature. Previous research has evidenced significant associations between single indicators of positivity and child and maternal outcomes (Hastings et al 2005b; Hastings, Beck and Hill, 2005a; Minnes, Perry & Weiss, 2015).

Exploring data from a national survey of caregivers raising a child with ID (Hastings, 2005a) Structural Equation Modelling (SEM) was used to investigate whether a latent construct of maternal positivity could be created using indicators that tapped onto multiple domains of maternal life. The secondary aim of this study was to explore whether a latent construct had significant relationships with maternal psychopathology, child behaviour problems and child prosocial behaviours which would indicate that maternal positivity could potentially function as a resilience variable.

Secondary data were also utilised in chapter 2. Ethical approval had been approved by Bangor University when data were initially collected, therefore, The University of Warwick did not require ethical review for a project involving secondary use of data. However ethical considerations were considered. For example, all data were anonymised before being released to myself by the original researcher and the outcomes of the analysis did not allow for participants to be identified in any way.

Chapter 2: The Construct of Maternal Positivity in Mothers of Children with Intellectual Disability[1]

[1.]This Chapter is based on: Jess, M., Totsika, V., & Hastings, R. P. (2017). The construct of maternal positivity in mothers of children with intellectual disability. *Journal of Intellectual Disability Research*, 61, 928-938. https://doi.org/10.1111/jir.12402

Abstract

Background: Despite the elevated levels of stress, anxiety and depression reported by mothers of children with intellectual disabilities (ID), these mothers also experience positive well-being and describe positive perceptions of their child. To date, maternal positivity has been operationalised in different ways using a variety of measures. In the present study, I tested whether a latent construct of maternal positivity could be derived from different measures of positivity.

Method: One hundred and thirty-five mothers of 89 boys and 46 girls with ID between 3 and 18 years of age completed measures on parental self-efficacy, their satisfaction with life, family satisfaction, their positive affect and their positive perceptions of their ID child. I conducted a confirmatory factor analysis of latent positivity, and subsequently tested its association with child social skills and behaviour problems, and maternal mental health.

Results: A latent maternal positivity factor achieved a statistically good fit using the five observed indicators of positivity. Parental self-efficacy had the strongest loading on the latent factor. Maternal positivity was significantly negatively associated with maternal psychological distress, maternal stress, and child problem behaviours and positively associated with child positive social behaviour.

Conclusions: These findings lend support to the importance of examining parental positivity in families of children with ID and using multiple indicators of positivity. Associations with negative psychological outcomes suggest that interventions focused on increasing parental positivity may have beneficial effects for parents. Further research is needed, especially in relation to such interventions.

There is clear evidence in research literature attesting to the difficulties and negative outcomes associated with raising a child with intellectual disabilities (ID). Compared to mothers of typically developing children, mothers raising children with ID report higher levels of parenting stress, anxiety and symptoms of depression (Eisenhower, Baker & Blacher, 2005; Estes et al, 2013; Olsson & Hwang, 2001). Children with ID tend to exhibit higher levels of behaviour problems compared with typically developing children and these behaviours very often explain the elevated parenting stress and negative mental health experienced by parents (Abbeduto et al, 2004; Glidden, Grein & Ludwig, 2014; Neece & Baker, 2008, Stores, Stores, Fellows & Buckley, 1998).

Although this negative impact is well documented throughout research literature, more recently it has become evident that parents of children with disabilities also experience positive mental health, positive perceptions, and report positive experiences (Hastings, 2016; Hastings & Taunt, 2002). For example, Totsika, Hastings, Emerson, Lancaster and Berridge (2011a) conducted a population-based cross-sectional comparison of mothers who had a child with an Autism Spectrum Disorder (ASD) only, ID only, combined ASD and ID and a child with neither condition (comparison group). The children were a representative sample of school age children in the UK. Mothers of children in the disability groups were found to exhibit higher levels of emotional disorder than mothers of the comparison group. However, there were no significant group differences in levels of maternal positive mental health. Similar findings were evident from a nationally representative sample of five year old children: Mothers of children with ID reported higher levels of serious mental ill-health than mothers of children without ID yet still reported similarly high levels of satisfaction with life (Totsika et al, 2011b). These data suggest that positive aspects of psychological well-being do not necessarily have to be compromised due to raising a child with ID, and that positive

indicators of well-being likely exist in parallel to poorer mental health in mothers of children with developmental disabilities (Hastings, 2016).

There has been a shift within disability research from focusing on negative outcomes such as stress and depression, to exploring positive outcomes (Bolourian & Blacher, 2016; Hastings, 2016; Stainton & Besser, 1998; Trute, Benzies & Worthington, 2012). For example, the Family Impact Questionnaire (FIQ; Donenberg & Baker, 1993) assesses parents' positive and negative perceptions of their child's impact on the family (compared to the impact of other children). Using the FIQ, Baker, Blacher, Crnic and Endelbrock (2002) found that mothers of three year old children with ID viewed their child as having a positive impact on their family. It could be argued that these findings were due to lesser demands exhibited in younger children or that challenging behaviours are easier to manage in early childhood. However, also using the FIQ, Blacher, Begum, Marcoulides and Baker (2013) reported similar findings in mothers of older children.

Several researchers have explicitly measured parental positive perceptions of children with ID using measures which are disability specific. For example, qualitative research conducted by Behr, Murphy and Summers (1992) led to the development of the Kansas Inventory of Parental Perceptions (KIPP) which is specifically for families of children with disabilities. The KIPP consists of four domains, of which one is the Positive Contributions Scale (PCS). The PCS was designed to identify the positive contributions children with disabilities make to their parents and their family.

Using the PCS, it has been identified that mothers of children with ID report positive perceptions of their child's contribution to their family and themselves. In a recent study, Vilaseca, Ferrer and Olmos (2014) found that mothers of children with ID between one and nineteen years old reported clinically significant levels of anxiety. However, Vilaseca and

colleagues (2014) also found that these mothers had a strong positive perception of their child which was not significantly associated with their anxiety. These findings suggested that mothers perceive their child with ID positively from early childhood to late adolescence, and further that positive constructs exist relatively independently from negative maternal outcomes even when these are at clinical levels.

Other researchers have found significant associations between parental positivity and psychological problems or distress in families of children with ID. For example, Hastings, Beck and Hill, (2005a) identified a small significant negative association between maternal positive perceptions of their child with ID and parenting stress and depression. Furthermore, in a recent study of mothers of children with ASD Kayfitz, Gragg and Orr (2010) found that maternal positive perceptions of their child were negatively associated with maternal parenting stress.

The relationship between challenging behaviour exhibited in children with disabilities and maternal mental health problems is well documented (e.g., Bromley, Hare, Davison, & Emerson, 2004; Gray et al, 2011; Hastings, 2002; Johnston et al, 2003; McConkey et al, 2008; Plant & Sanders, 2007). However, the relationship between indicators of positivity and child behaviour problems has received less research attention. The limited research that does exist suggests that child problem behaviours relate with positive indicators very much in the same way that poor maternal mental health does (i.e., there is a negative association) (Crnic, Gaze & Hoffman, 2005; Suldo & Heubner, 2005; Totsika et al., 2013). In addition to positive perceptions and impact, researchers have considered the role of parental feelings of self-efficacy as a single indicator of positivity. In the context of childhood disability, a parent who is positive about his/her parenting efficacy is likely to be confident in dealing with and perceive to be in control of their child's behaviour problems. Existing research studies have

generally found this predicted association (Jones & Prinz, 2005; Lloyd & Hastings, 2009; Sanders & Woolley, 2004).

Much less attention has been paid to the positive social behaviours than the behaviour problems of children with ID. One might expect positive child behaviours to have a positive association with positive parental outcomes. Prosocial/positive behaviours include turn taking, sharing and compliance with adult instructions. Although prosocial behaviours are exhibited less in children with ID compared to typically developing children, evidence indicates that parents of children with higher levels of prosocial behaviours report higher scores on individual indicators of positivity including positive perceptions and parental efficacy (Lloyd & Hastings, 2009).

In ID family research with a focus on parental positivity, researchers have used a variety of single measures of positivity including general positive constructs (life satisfaction, family satisfaction, and positive affect: Ekas, Lickenbrock & Whitman, 2010; Hassall, Rose & McDonald, 2005; Lloyd & Hastings 2009), and disability specific measures such as the PCS and the Positive Gain Scale (Blacher & Baker, 2007; Cianfaglione et al., 2015; Jones, Totsika, Hastings, Petalas, 2013; Weiss & Lunsky, 2011)

However, to date the associations between single indicators of positivity and child and maternal outcomes have been relatively small in comparable cross-sectional studies with correlation coefficients either near zero (e.g., $r = -.02$; Positive Contributions and child behaviour problems; Hastings et al, 2005b) or small (e.g., $r = -.29$; Positive Gains and Parental Distress; Minnes, Perry & Weiss, 2015). It is possible that positive and negative constructs are not closely related (i.e., are relatively independent). However, researchers have also not explored the relationships between different indicators of parental positivity in ID family research. In addition, there is no consensus on an overall construct of parental

positivity. Given that some indicators of positivity used in ID research have been disability-specific (KIPP-PC; Behr et al, 1992; Donenberg & Baker, 1993; PGS: Pit-ten Cate, 2003) and others more general, it is important to explore whether these represent one underlying positivity construct or distinct domains.

The primary aim of the present study was to investigate whether it was possible to describe maternal positivity by developing a latent construct drawing on several indicators of positivity. Given previous research findings concerning the relationship between single indicators of positivity and child and maternal outcomes, our secondary aim was to clarify whether the association between a latent construct of maternal positivity and maternal negative psychological outcomes and maternal positivity and child behaviours would follow the same direction of associations as previous research on single indicators of positivity and child and maternal outcomes. The main questions to be addressed in this study were, 'Can a latent construct of Maternal Positivity be created using multiple single indicators of positivity?' and 'Would a latent construct of Maternal Positivity have a significant relationship with child and maternal outcomes?' Importantly, identifying a negative relationship between a latent construct of maternal positivity and maternal psychopathology would suggest that maternal positivity may function as a resilience variable.

Method

Participants

One hundred and thirty five mothers of children with ID participated in the research. Their ages ranged from 23 years to 57 years (*M*=39.45 years, *SD*=7.23). A majority of the mothers were married or living with a partner (*n*=102), although 33 (24.44%) were divorced. The mothers in the sample were well educated: 68 (50.37%) had a college or university education, 47 (34.81%) had secondary school leaving qualifications, and 20 (14.81%) mothers had no

formal educational qualifications. Sixty-five mothers (48.15%) had paid work outside the home and the remaining 70 (51.85%) mothers were not in paid employment. Of the 65 mothers who were in paid employment, 18 (27.69%) worked full-time and 47 (72.31%) worked part-time.

The children with ID were 89 (65.93%) boys and 46 (34.07%) girls. Their ages ranged from 3 years to 18 years (*M*=10.02 years; *SD*= 4.11 years). Fifty-five (40.74%) children were reported as having a diagnosis of Autism in addition to ID, 25 (18.52%) had Down Syndrome, 16 (11.85%) had Cerebral Palsy, and the remainder were a mixed aetiology ID group. The diagnoses were based on parental reports, and I did not have access to clinical notes to establish the validity of these reports. At the time of data collection, all the children attended Special Schools in North Wales or the North West of England in which primarily children with severe intellectual disability were educated. The majority of households had a total of 1 (22.22%) or 2 (42.22%) children living at home. Thirty five households had 3 (25.93%) children at home, nine had 4 (6.67%) children and 3 had 5 (2.22%) children. One mother did not report on the total number of children living in the family home.

Measures

A total of nine measures were used in this study, in addition to a demographic questionnaire that assessed sociodemographic characteristics reported in the participants' section.

Maternal Positivity measures

Positivity data was collected from five measures: three general positive measures and two focused on positivity in the context of parenting the child with ID.

The Satisfaction with Life Scale (Diener, Emmons, Larsen & Griffin, 1985) (see Appendix II) is a five- item scale that asks participants to report their degree of agreement or disagreement to statements such as, "The conditions of my life are excellent" on a seven-

point Likert-type scale. This scale was designed to measure subjective well-being among normative populations but has been used successfully and shown to have excellent psychometric properties when used with mothers of children with ID (Griffith, Hastings, Nash & Hill, 2010). Cronbach's alpha for the present sample was .87.

The Family Satisfaction Scale has fourteen items and measures family cohesion and adaptability (Olson & Wilson, 1982) (see Appendix III). Participants are asked to report their degree of satisfaction to statements such as, "How satisfied are you with the amount of time you spend together as a family?" and "How satisfied are you with how often you make decisions as a family, rather than individually?" This scale was modified to be used by parents with a dependant. Therefore, two items were excluded ("How satisfied are you with how often parents make decisions in your family?" And "How satisfied are you with how much mother and father argue with each other?") as these items reflected the satisfaction of a dependent child. This scale was designed for a normative population but has been used successfully and shown to have good levels of reliability when used with mothers of children with ID (Griffith et al., 2010). Cronbach's alpha for the present sample was .94.

The Positive Affect Scale taken from the Positive and Negative Affect Scales (Watson et al. 1988) (see Appendix IV) measures positive feelings and emotions, by asking mothers to rate 10 words such as "Interested" and "Determined." Mothers rated to what extent each word applied to them at the time of completion on a Likert-type scale ranging from "very slight or not at all" to "extremely." This scale was designed for a normative population but has been used successfully and shown to have good levels of reliability when used with mothers of children with ID (Hastings et al, 2005a). Cronbach's alpha for the present sample was .91.

The first of the positivity measures focused on experiences of parenting the child with ID was the Parental Self-Efficacy Scale (Hastings & Brown, 2002) (see Appendix V). This measure consisted of five efficacy items (e.g. feelings of confidence in parenting, a rating of how difficult they find it to parent their child with ID). Items are rated from "not at all" to "very". The Cronbach's alpha for the present sample was .89. The Positive Contributions Scale from the Kansas Inventory of Parental Perceptions (PCS; Behr et al.1992) was used to measure mothers' perceptions of the positive contributions their child with a disability has brought to themselves (such as, personal growth and maturity, happiness and fulfilment), to the wider family (strength and family closeness) and that the child has a number of positive characteristics (such as, kind and loving). This scale was developed for parents of children with ID and has been used successfully demonstrating good reliability with mothers of children with ID (Lloyd & Hastings, 2008). Cronbach's alpha for the total score on the PCS for the present sample was .93. Descriptive data for single indicators of positivity are displayed in Table 2.1

Table 2.1. Descriptive data for Maternal Positivity variables

	Parental self-efficacy	Satisfaction with Life	Family Satisfaction	Positive Affect	KIPP-PC
Total	135	135	135	135	135
Mean Score (range; SD)	24.65 (5-35; 5.79)	20.51 (5-35; 7.2)	37.37 (12-60; 9.98)	33.08 (10-49; 7.92)	135 (87-183; 18.97)

Maternal psychological problems measures

Two measures of mothers' psychological problems were included. Maternal stress, related to having a child with a disability in the family was measured using the Parent and Family Problems sub-scale of the Questionnaire on Resources and Stress- Friedrich short form (QRS-F; Friedrich, Greenberg & Crnic, 1983) (see Appendix VII). Five items were excluded as they have been identified as a robust measure of depression (Glidden & Floyd, 1997). This

was to ensure that there was no overlap between the measures of stress and of mental health problems used in the present research. The QRS-F was designed for families of children with disability and has good reliability when used with mothers of children with ID (Griffith et al., 2011). A Kuder-Richardson coefficient of .84 was gained for the present sample for the total parent and family problems score.

Maternal psychological distress was assessed using the Hospital Anxiety and Depression Scales (HADS; Zigmond and Snaith, 1983). This consists of two, seven-item subscales that are rated from zero to three that measure levels of anxiety and depression. A dimensional approach was taken for the main analyses, with a total score of the two sub-scales being used. Combining scores of both scales is a method to obtain a general measure of psychological distress (Crawford, Henry, Crombie & Taylor, 2001). A total score of more than 22 indicates moderate to severe cases of psychological distress. The HADS was developed to be used in a medical outpatient clinic but has been widely used in samples of parents of children with disabilities (Beck, Hastings, Daley & Stevenson, 2004; MacDonald, Hastings & Fitzsimmons, 2010). The Cronbach's alpha for the psychological distress total score for the present sample was .87. Descriptive data for child and maternal outcome measures are displayed in Table 2.2.

Table 2.2. Descriptive data for Child and Maternal outcome measures

	HADS	QRS-F	NCBRF	Reiss
Total	135	132	132	132
Mean Score (range; SD)	14.49 (0-39; 6.74)	22.09 (2-41; 8.05)	11.89 (1,30; 5.67)	24.17 (0-76; 18.18)

Child behaviour measures

The Reiss Scales for Children's Dual Diagnosis (Reiss & Valenti-Hein, 1994) is a 60-item

measure designed to assess psychopathology in children with intellectual disabilities. Each

item is scored on a three-point scale, "No Problem", "Problem", or "Major Problem". There

are 10 subscales (attention deficit, anger, anxiety, conduct disorder, depression, autism,

psychosis, self-esteem, somatoform and withdrawn behaviours). These scales can be used

separately or summed to form a total behaviour problem score and this has been used in

several studies of children with ID (Lloyd & Hastings, 2009; Maes, Broekman, Došen, Nauts,

2003). For the present study I used the total score only. The Cronbach's alpha for the total

score in the present sample was.95.

The Nisonger Child Behaviour Rating Form (NCBRF; Aman, Tassé, Rojahn &

Hammer, 1996) (see Appendix VII) is a 76-item measure consisting of two scales designed to

assess several different behaviours in children with ID. The social competence scale of the

NCBRF was used to measure child positive social behaviour. This is a ten-item scale of

positive behaviours that are described as either calm/compliant (e.g., followed rules) or

adaptive/ social behaviours (e.g., participated in group activities). Items are rated from "not

true" to "completely always true". This measure has been used successfully with ID children

(Waltz & Benson, 2002) The Cronbach's alpha for the total child positive social behaviour

score for the present sample was .88.

Procedure

Participants were a sub-sample from a study of families of children with ID (Hastings et al.,

2005a). Families were recruited through the child's special school. Information packs about

the research were sent to families via their children who attended a school for children with

ID. Within the information pack was a response form and a business reply envelope. When

response forms had been returned, separate questionnaire packs and consent forms were

posted to the primary caregiver (mother). Families were offered a small payment for

returning the questionnaires to recognise the time they had spent participating in the research.

Statistical Analysis

I conducted the analyses with structural equation modelling (SEM) using AMOS 22

(Arbuckle, 2013). Analyses were conducted in two distinct phases. I conducted a

Confirmatory Factor Analysis (CFA) to test the hypothesis that a latent construct of maternal

positivity could be generated from the five indicators of positivity. I then explored

associations between maternal positivity and maternal mental health problems and

associations between maternal positivity and child behaviour (behaviour problems and

positive social behaviour) in 4 separate SEM models. To evaluate model fit I used several

criteria: the ratio of chi-square to degrees of freedom (CMIN/DF) under 2 (Tabachnick &

Fidell, 2007), Root Mean Square Error of Approximation (RMSEA) under .05 (Browne &

Cudeck, 1993) and the Comparative Fit Index (CFI) above .95 (Hu & Bentler, 1999).

Results

Developing the Latent Construct of Maternal Positivity

I initially ran bivariate correlations between each of the measures of maternal positivity. I

then fitted the five indicators without correlating any of their error terms. However, the fit

indices exceeded the values that were used as guidance for a good fit (see earlier). Based on

the strength of the bivariate correlations, I selected error terms that were allowed to correlate

between indicators (depicted by a double-headed arrow in the model in Figure 2.1). The

model with the correlated errors represented an improved fit to the data as supported by the

fit indices (see Table 2.4). In addition, I used the Akaike Information Criterion (AIC) which

is used in the comparison of two or more nested models, with smaller values representing a

better fit of the hypothesised model (Hu & Bentler, 1995). As can be seen in Table 2.4, the

AIC also indicates the better fit of the model with correlated error terms.

Figure 2.1. The latent construct of maternal positivity and the factor loadings of the 5 indicators of positivity.
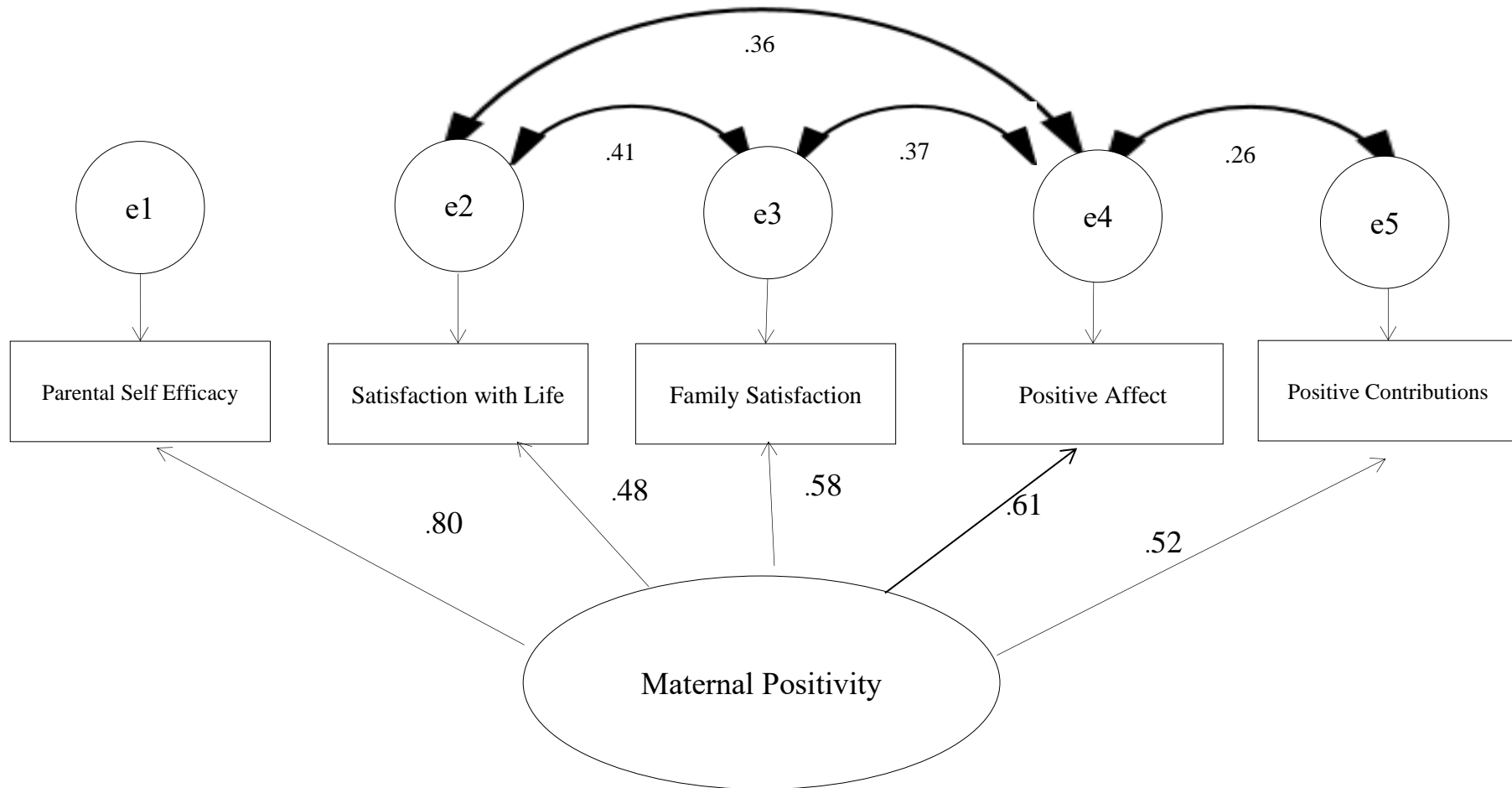
Table 2.3. Associations (Pearson's correlation Coefficient) between child behaviour and maternal measures.

Measure	1	2	3	4	5	6	7	8	9
1.Parental Stress	-	-.53**	-.52**	-.46**	-.41**	-.33**	-.54**	.56**	.50**
2.Parental Self Efficacy		-	.45**	.46**	.40**	.37**	.38**	-.42**	-.46**
3.Satisfaction with Life			-	.62**	.54**	.24**	.20*	-.26**	-.56**
4.Family Satisfaction				-	.57**	.33**	.27**	-.35**	-.64**
5.Positive Affect					-	.45**	.30**	-.31**	-.53**
6. Positive Contributions						-	.28**	-.26**	-.13
7.Child Positive Social Behaviours							-	-.55**	-.32**
8.Child Behaviour Problems								-	.47**
9. Psychological Distress									-

** Correlation is at the 0.01 level (2-tailed),* Correlation is at the 0.05 level (2-tailed)

Table 2.4. Model fit indices for the latent constructs of maternal positivity.

	χ^2	p-value	CMIN/DF	RMSEA	CFI	AIC
Uncorrelated errors terms	13.77	.02	2.76	.11	.96	43.77
Correlated error terms	1.33	.25	1.33	.05	1	39.33

Table 2.5. Model fit indices for associations between the latent construct of maternal positivity and child and maternal outcomes.

Model	χ^2	p-value	CMIN/DF	RMSEA	CFI	AIC
Maternal Positivity x Parental Stress	3.91	.56	.78	<.001	1	47.91
Maternal Positivity x Psychological Distress	19.09	.002	3.82	.14	.95	63.09
Maternal Positivity x Child Prosocial Behaviours	4.15	.53	.83	<.001	1	48.15
Maternal Positivity x Child Behaviour Problems	3.15	.63	.63	<.001	1	47.15

In the final model (Figure 2.1), the factor loadings of the five indicators were all significant ($p <$.001) with Parental Efficacy having the strongest factor loading, β=.80 and thus the strongest contribution to the latent positivity construct.

Associations between Maternal Positivity and other maternal/child variables

I examined associations between the maternal positivity latent construct and other study variables. Maternal positivity had a significant negative association with maternal parenting stress ($\beta = -.74$, $p= <$.001), a significant negative association with maternal psychological distress ($\beta = -.76$, $p = .006$), a significant positive association with child positive social

behaviour (β= .48, p <.001), and a significant negative association with children's behaviour problems (β= - .54, p <.001.) See Table 2.5 for model fit indices.

Discussion

In the present study, I tested the potential of fitting a latent maternal positivity factor using five indicators with some parenting or disability-specific (parenting self-efficacy, positive contribution) and others representing general positivity (family satisfaction, life satisfaction, positive affect). Findings suggested that there is indeed an underlying positivity construct that can be described using general positive measures as well as positivity measures specifically focused on the experience of parenting a child with ID. Parenting Self-Efficacy loaded the strongest to the construct of maternal positivity and perceived Positive Contributions loaded the weakest.

In the second stage of analyses, it was found that the latent construct of maternal positivity had a significant positive association with child positive social behaviour, and negative associations with maternal psychological distress, parental stress and child behaviour problems. These associations were reasonably substantial, suggesting robust relationships with both measures of maternal mental health and child functioning. These findings are in contrast to much of the previous research suggesting weak relationships between single indicators of positivity and both child functioning and maternal psychological problems (stress, anxiety and depression).

Importantly I was able to replicate the negative associations, identified in previous studies, between single positivity indicators and child behaviour problems and maternal mental health problems using the latent construct of maternal positivity in place of a single

indicator. Furthermore, I added to the extant literature by finding that positive child behaviours were positivity associated with maternal positivity.

In this study, single indicators of maternal positivity had positive associations with one another (see Table 2.3), theoretically increasing the overall strength of maternal positivity, thus explaining why the construct of maternal positivity has a stronger association with child and maternal outcomes than single indicators. Not only are single indicators not broad enough to fully represent maternal positivity in its entirety but isolated, they do not have particularly strong associations with child and maternal outcomes.

There is a need for mothers to find effective ways to reduce the levels of stress and other negative outcomes associated with raising a child with ID. The current theoretical findings indicate that maternal positivity could be conceptualised as a resilience variable in that it functions by reducing negative maternal outcomes. Thus, findings from this study have practical implications for targeting key constructs for intervention. Again, longitudinal data are required to confirm the findings. However, the findings suggest that targeting parental positivity (especially mothers' feelings of efficacy in the parenting role) may help to reduce maternal psychological problems and potentially also improve child functioning. These suggestions are borne out by results from existing intervention studies. For example, Hudson and colleagues (2003) found that intervention methods which increased feelings of parental efficacy in families of children with ID led to a reduction in child behaviour problems and parental stress.

It is important that a number of methodological limitations of the current study are considered. First, diagnostic status was not confirmed by a practitioner or clinical reports. However, the children were all attending specialist schools and so were likely to have clinically diagnosed ID. Second, mothers completed all measures. Therefore, there is a

problem of source variance that may have inflated associations between study variables. Future research studies should incorporate multi-informant measures especially of child functioning or independent measures of maternal well-being perhaps incorporating observations of happiness for example.

Importantly, conclusions based on latent variables cannot be generalised and are only specific to participants in the study. Therefore, it is worth considering that a different set of positivity indicators may be more applicable to mothers of children with an ID diagnosis not included in this study. In addition, different associations between maternal positivity and outcome measures may have emerged for a different group of mothers.

As suggested earlier, due to the cross-sectional study design, causality cannot be inferred and there is a clear need for longitudinal studies of parental positivity building on multiple indicators of positivity. Further research should also investigate which indicators make up paternal positivity and whether such a construct would have similar associations with child and paternal outcomes in an effort to support fathers raising children with ID.

Orientation to Chapter 3

The findings from chapter 2 suggest that a construct of maternal positivity for mothers raising children with ID can be derived from individual indicators of general positivity, positivity specific to parenting and positivity specific to raising a child with a disability. Furthermore, chapter 2 evidenced that maternal positivity as a latent construct is experienced independently of maternal psychopathology of which there was a significantly negative association.

Considering these findings, the function of maternal positivity within the context of resilience was examined. Families of children with ID face many challenges associated with their child however whilst they may struggle most families do well. Resilience theory proposes that resilience is the ability to thrive in the face of adversity and that resilience variables have one of two functions; compensatory and/ or protective (Luthar, 1991). A compensatory function has a direct main effect and a protective function buffers the impact of the risk or adversity. Therefore, if maternal positivity did function as a resilience variable, in accordance to resilience theory, it would be expected that maternal positivity would have a direct impact on maternal outcomes, for example by reducing levels of maternal stress and depression. Alternatively, maternal positivity could also be conceptualised as a resilience variable if it buffered the negative impact of a stressor (i.e. child behaviour problems) on maternal well-being outcomes.

The aim of this study was to determine whether the multi-indicator latent construct of maternal positivity functioned as a resilience variable cross-sectionally and/or longitudinally.

Data for chapter 3 were drawn from a previous database (Hastings, Beck & Hill; 2005). The University of Warwick did not require ethical review for a project involving secondary use of data. However, ethical considerations were made. For example, all data were anonymised

before being released to myself by the original researcher and the outcomes of the analysis

did not allow for participants to be identified in any way.

Chapter 3: Maternal Stress and the Functions of Positivity in Mothers of Children with Intellectual Disability[1]

Abstract

Background: Although mothers raising children with Intellectual Disability (ID) report poorer mental health than parents raising typically developing children, they also report feelings of positivity; both generally and specific to their child. To date little is known about the function of maternal positivity thus, the aim of this study was to investigate the putative compensatory and protective functions of a latent construct of maternal positivity, within both a cross-sectional and one- year longitudinal framework.

Method: Participants included 135 mothers of children with severe ID who were between 3 and 18 years of age. Multiple linear regression models investigated the potential function of maternal positivity when child behaviour problems and child psychopathology were conceptualised as risk variables. Maternal psychopathology and parenting stress were framed as outcome variables.

Results: At a cross-sectional level, maternal positivity was found to be a significant independent predictor of maternal stress and moderated the impact of child behaviour problems on maternal parenting stress. Longitudinally, maternal positivity did not have a direct effect on later parenting stress nor function as a moderator.

Conclusions: Findings from the cross-sectional analysis are consistent with the view that positivity serves a compensatory function. Further exploration is needed to understand the longitudinal function of maternal positivity.

High quality population-based research data suggest that mothers raising children with intellectual disabilities (ID) experience higher levels of stress and mental health problems compared to other mothers (Emerson, 2003; Emerson & Llewellyn, 2008; Totsika et al, 2011a). Despite the difficulties and challenges, many parents of children with ID are able to thrive and express a positive attitude towards life (Blacher & Baker, 2007; Gardner & Harmon, 2002) and their child (Hastings & Taunt, 2002; Hastings et al, 2002). Thus, the question of whether parents are affected negatively by raising a child with ID involves a complex answer: they do face more stress, but they also report significant positive outcomes and positive well-being often to the same extent as do other parents (Hastings, 2016).

There has been an increase of research focussing on positive constructs and outcomes for parents of children with ID, particularly mothers. In a majority of cases the primary carer of children in any family is the mother. Therefore, any adverse or beneficial effects associated with raising a child would arguably have a greater salience for the mother - more so than for other family members. Lloyd and Hastings (2009) explored hope (defined as one's perceived ability to reach a goal) and its relationship with parental well-being. It was found that mothers of children with ID who reported higher levels of hope reported lower levels of anxiety, depression and stress. Other positive constructs such as life satisfaction and positive affect have also been found to have negative associations with parenting stress and depression (Ekas et al, 2010; Lloyd & Hastings, 2009).

Disability-specific measures of positive experiences have been developed for parents of children with ID, including the Positive Gain Scale (Griffith et al, 2011; Jones et al, 2014; MacDonald et al, 2010; Pit-ten Cate, 2003; Weiss et al, 2015), the Positive Contributions Scale (Behr et al, 1992; Hastings et al, 2002; Hastings et al, 2005a; Vilaseca et al, 2014) and the Positive Impact scale of the Family Impact Questionnaire (FIQ; Donenberg & Baker, 1993). Evidence suggests not only that positivity exists within these families but in some

instances children with ID may have a more positive impact on their family than typically

developing children have on theirs (Blacher et al, 2013). In addition, existing research

indicates that such positive constructs often have a distinct inverse relationship with negative

outcome measures (Hastings et al, 2005b; Lloyd & Hastings, 2008; Minnes et al, 2015;

Vilaseca et al, 2014).

Less research attention has been given to the putative functions of positive

perceptions and positive functioning for parents of children with ID. In an early review of

parental positivity in developmental disabilities, Hastings and Taunt (2002) drew on risk and

resilience theories (Fraser, Richman & Galinsky, 1999; Luthar, 1991; Luthar & Zigler, 1991;

Rutter, 1985) to identify potential different functions of positivity. Resilience has been

described as the ability to thrive despite exposure to adversity or stressful life events (Luthar,

1991), and as the ability to withstand stress (Heiman, 2002). In ID research, a resilience

perspective is related to the considerable external and child-related challenges when raising a

child with ID and the fact that many parents and families thrive despite these stressors. To be

able to consider the functions of positivity, it is also necessary first to clarify a reliable

stressor for parents of children with ID. Stressors would ideally be evidenced as risk factors,

variables causally related to parental well-being. The behaviour problems of children with ID

is the most consistently identified risk factor for poorer parental well-being in families of

children with ID. Several longitudinal research studies show that child behaviour problems

are a significant predictor of later poorer parental mental health (Baker et al, 2003; Herring et

al, 2006; Lounds et al, 2007; Neece & Baker, 2008; Neece et al, 2012).

Luthar (1991) outlined resilience variables as serving two distinct functions:

compensatory and protective. Variables which function as protective moderate the effects of

life stressors. Protective factors improve outcomes in the face of stressors (i.e., risk) but not

necessarily otherwise. Within the context of ID research, the stressor, or risk variable could

be child behaviour problems. Positive constructs would have a protective function if when exposed to high levels of child behaviour problems those with high levels of positivity were less affected in terms of their mental health than those with lower levels of positivity. A compensatory function is a main effect as opposed to an interactive/moderating effect. In the current case, positivity would serve a compensatory function if high levels of positivity predicted better maternal mental health. However, this relationship would be independent of any association with child behaviour problems as a risk factor. In a recent cross-sectional study Halstead and colleagues (2018) explored whether maternal resilience, defined by The Brief Resilience Coping Scale (Sinclair & Wallston, 2004), had either a protective or compensatory function for mothers of children with ID when child emotional and behavioural problems functioned as risk factors. The study found strong support for a compensatory function, with maternal resilience having a direct effect on maternal anxiety, depression and parenting stress. However, maternal resilience was found to have only a slight protective function for maternal stress only thus it was concluded that higher levels of resilience were associated with better maternal outcomes.

Although not directly referring to either compensatory or protective effects, existing research has examined both of these potential functions within single indicators of parental positivity in families of children with developmental disabilities. The interest in maternal positivity in these families brings together two perspectives. The first is a theoretical orientation towards strengths-based approaches in ID research (Wehmeyer et al., 2017). This approach advocates a research focus on strengths, which is consistent with cumulative evidence that families of children with ID can experience positive adaptation, despite any negative outcomes (Hastings, 2016) The second perspective that informed the focus of this study was the evidence from family research in typical development that different dimensions of parenting or parental well-being (positive and negative) are correlated with different

outcomes in children (Anthony et al, 2005; Benzies et al, 2004; Hautmann et al, 2015; Morgan et al, 2002; Salari et al, 2014). Similarly, current evidence in ID research supports a small positive association between positive parenting and child outcomes (Dyches et al., 2012), but it is unclear how maternal positivity (i.e., a psychological state of positive orientation) is associated with other aspects of maternal mental health and also child outcomes in this population. For example, there is a negative association between maternal self-efficacy (one of the aspects of the positivity construct, as detailed below) and maternal mental health problems (Hassall et al, 2005; Hastings & Brown, 2002; Kuhn & Carter, 2006). In terms of a putative protective function for parental positivity, Weiss, MacMullin and Lunsky (2015) found that high levels of child aggression in children with Autism Spectrum Disorder (ASD) were not related to distress in mothers who also reported high rates of positive gain. Previous cross-sectional studies of families raising children with ID and or ASD have also identified different variables that could be considered indicators of positivity, such as positive impact, and positive reappraisal coping styles that have moderated the relationship between stressors and parental mental health (Blacher & Baker, 2007; Dunn et al, 2001; Glidden et al, 2006; Lyons et al, 2010).

Existing research on the function of parental positivity has been largely limited to cross-sectional studies. Even within longitudinal studies, the prospective nature of the available data has not been reported. For example, parental optimism moderated the relationship between child behaviour problems and parental depression (primarily for mothers) in families of young children with ID when the child was three years old and also when the child was four years old (Baker et al, 2005). Although this study demonstrates that optimism functioned as a moderator at two time points within the same sample, the results do not determine whether parental optimism, when the child was three, would moderate the impact of child behaviour problems (at three years old) on later parental depression (one year

later). Thus, the function of optimism over time was not explored. To fully examine either a compensatory or protective function, prospective research designs are needed: positivity may reduce later negative outcomes, or moderate current exposure to risk in terms of later outcomes.

A further methodological limitation of parental positivity research to date is the lack of clear definition and measurement of positivity. In a previous cross-sectional study, myswlf and colleagues had explored the dimensions of this construct (Jess, Totsika & Hastings, 2017). Using Confirmatory Factor Analysis (CFA), I created a latent construct of maternal positivity from five single indicators (see figure 2.1): Parental Self-Efficacy (Hastings & Brown, 2002); parental general Satisfaction with Life (Diener et al, 1985); Family Satisfaction (Olson & Wilson, 1982); general Positive Affect (Watson & Clark, 1988); and Positive Perceptions of their child with ID (the Positive Contributions Scale: Behr et al, 1992) (see Appendices II-VI). I used several criteria to evaluate model fit: the ratio of chi-square to degrees of freedom (CMIN/DF) under 2 (Tabachnick & Fidell, 2007), Root Mean Square Error of Approximation (RMSEA) under .05 (Browne & Cudeck, 1993), and the Comparative Fit Index (CFI) above .95 (Hu & Bentler, 1999). A confirmatory factor analysis indicated that these five indicators produced a well-fitting construct of positivity (χ^2/df ratio=1.33, CFI= 1.00, and RMSEA= .05). Thus, one latent variable was created from five single indicators of positivity, which was then defined as 'maternal positivity'. Furthermore, it was found that the latent variable of maternal positivity was negatively associated with maternal psychological distress and parenting stress, further confirming the construct's validity.

Although in the previous study I identified a negative association between maternal positivity and poor maternal mental health outcomes, the functions of maternal positivity were not examined. Therefore, the aim of the current study was to explore the function of

maternal positivity both cross-sectionally and longitudinally. Measures of children's behaviour and mental health problems were included as putative risk factors for maternal psychological adjustment. I examined both protective and compensatory functions (as defined earlier) for maternal positivity. In the absence of existing research, specific hypotheses were not examined. However, considering risk and resilience theories and the prospect that maternal positivity may function as a resilience variable given it's demonstrated relationship with maternal psychopathology in the previous chapter, the research question to be addressed in this study was, 'Does Maternal Positivity have a protective or a compensatory function either cross-sectionally and/ or longitudinally?'

Method

Participants

At Time 1 Participants were 135 mothers of children with severe ID (see Table 3.1). Their ages ranged from 23 years to 57 years (*M*=39.45 years, *SD*=7.23). A majority of the mothers were married or living with a partner (*n*=102), although 33 (24.4%) were divorced. The mothers were well educated: 68 (50.4%) had a college or university education, 47 (34.8%) had secondary school leaving qualifications, and 20 (14.8%) mothers had no formal educational qualifications. Sixty-five mothers (48.1%) had paid work outside the home and the remaining 70 (51.9%) mothers were not in paid employment. Of the 65 mothers who were in paid employment, 18 (13.3%) worked full-time and 47 (34.8%) worked part-time. The majority of households had a total of one (22.2%) or two (42.2%) children living at home. Thirty-five households had three (25.9%) children at home, nine had four (6.7%), and three had five (2.2%) children living at home. One mother did not report on the total number of children living in the family home.

There were 89 (65.9%) boys and 46 (34.1%) girls with ID, and diagnoses were based on parental reports. Fifty-five (40.7%) children were reported as having an additional

diagnosis of Autism, 25 (18.5%) had Down's syndrome, 16 (11.9%) had Cerebral Palsy, and 39 were a mixed aetiology ID group (28.9%). Children's ages ranged from 3 to 18 years (*M*=10.02 years; *SD*= 4.11 years).

At Time 2 at one year follow-up 110 of the original 135 mothers participated. The follow-up sample were very similar to the original sample in terms of demographic characteristics (see Table 3.1).

Table 3.1. Demographic characteristics of mothers and children at Times 1 and 2

Variable	Time 1	Time 2
	N (%)	**N (%)**
Total number of mothers	135	110
Mean age of mothers (range; SD)	39.45 (23-57; 7.23)	39.92 (23-57; 7.23)
Married	86 (63.7%)	73 (66.4%)
Living with partner	16 (11.9%)	13 (11.8%)
Divorced	33 (24.4%)	24 (21.8%)
University or college education	68 (50.4%)	57 (51.8%)
Secondary school qualifications	47 (34.8%)	42 (38.2%)
No formal education qualifications	20 (14.8%)	11 (10%)
Employment outside home	65 (48.1%)	54 (49.1%)
Not in employment	70 (51.9%)	56 (50.9%)

Full time employment	18 (13.3%)	16 (14.5%)
Part time employment	47 (34.8%)	38 (34.5%)
Mean number of children in household (range; SD)	2.2 (1-5; .95)	2.3 (1-5; 2.28)
Mean age of children (range; SD)	10.02 (3-18; 4.11)	9.94 (3-18; 4.16)
Girls	46 (34.1%)	37 (33.6%)
Boys	89 (65.9%)	73 (66.4%)
Autism + ID	55 (40.7%)	42 (38.2%)
Down's Syndrome	25 (18.5%)	23 (20.9%)
Cerebral Palsy	16 (11.9%)	15 (13.6%)
Mixed Aetiology	39 (28.9%)	30 (27.3%)

Procedure

The mothers included in the present study were those from a cross-sectional study (Hastings et al., 2005a; Jess, Totsika & Hastings, 2018) who completed measures at Time 1 and 110 of those mothers completed follow-up measures 12 months after the initial data collection. All measures described below were gathered at the first data collection point (including the five positivity indicators contributing to the latent positivity construct). Maternal parenting stress and psychological distress (HADS) were also gathered at the first point of data collection and after 12 months had elapsed (Time 2).

Families were recruited through special schools for children with severe ID. Letters and information packs about the research were sent to more than 50 schools. Within the information pack was a response form and a paid reply envelope. Reply slips were received from 188 mothers and 72 fathers. Reply slips did not include information about the name of the child's school.

When response forms had been returned, separate questionnaire packs and consent forms were mailed to the primary caregiver. Families were offered a small payment for returning the questionnaires to recognise the time they had spent participating in the research. One year after the initial data collection, the families who took part at Time 1 were re-contacted to provide follow-up data.

Measures

In total, five measures were used in this study. In addition, a demographic questionnaire that identified sociodemographic characteristics reported in the Participants section was included.

The Behavior Problems Inventory (BPI-01; Rojahn et al; 2001) was used to measure child behaviour problems or "challenging behaviour". The BPI-01 is a 52-item instrument that measures self-injurious, stereotypic and aggressive behaviours in individuals with developmental disabilities. Self-injurious behaviours are defined as behaviours which can cause damage to the subject's own body (Rojahn et al, 2001) such as hitting of the head or other body parts. Stereotypic behaviours are repeated body movements that are not part of a goal-directed act such as rocking and twirling and/ or smelling objects. Aggressive or destructive behaviours are abusive deliberate attacks against other individuals or objects (Rojahn et al.; 2001). This measure has two response scales, frequency and severity. For this study only the frequency scale was used. Measuring frequency of child behaviour problems is arguably more relevant to the experience of raising a child with ID given that child behaviour

problems are known to occur more frequently in children with ID than they do in children without disabilities, whether the behaviours are high or low in severity. Furthermore, there is evidence to suggest that reporting on frequency shows greater stability than when participants report on intensity terms (Krabbe & Forkmann, 2012). Therefore, I decided it to be more advantageous to report on frequency scales than it would be to report on intensity. Each item is scored on a five-point frequency scale, "never", "monthly", "weekly", "daily" and "hourly" ranging from a score of 0 (never) to 4 (hourly). Higher scores represent higher frequency. The alpha coefficient for the total BPI-01 frequency score was .94 in the present study.

The Reiss Scales for Children's Dual Diagnosis (Reiss & Valenti-Hein, 1994) were used to measure children's behaviour problems and psychopathology as indicative of mental health difficulties. This is a 60-item measure designed to assess mental health in children with ID. Each item is scored on a three-point scale, "No Problem", "Problem", or "Major Problem". There are 10 subscales (attention deficit, anger, anxiety, conduct disorder, depression, autism, psychosis, self-esteem, somatoform and withdrawn behaviours). These scales can be used separately or summed to form a total problems score. For the present study, the total problems score was used. The Cronbach's alpha for the total score in the present sample was .95.

Maternal positivity was a latent variable constructed using five indicators of positivity (see Introduction; Jess, Totsika & Hastings, 2018) (see figure 2.1). It was designed to measure overall positivity in mothers of children with ID and is comprised of disability-specific, parenting specific and general measures of positivity. High scores indicate higher levels of positivity and low scores indicate lower levels of positivity. Sample items include: "The conditions of my life are excellent" (Satisfaction with Life Scale) (see Appendix II), "How satisfied are you with the amount of time you spend together as a family?" (Family Satisfaction Scale) (see Appendix III), "I consider my child to be the reason I am more productive" (Positive

Contributions Scale), "How confident are you in parenting your child with special needs?" (Parenting Efficacy) (see Appendix V) and "Indicate to what extent you feel enthusiastic at this present moment" (Positive Affect Scale) (see Appendix IV). Estimated regression-based factor scores for maternal positivity were extracted from AMOS 22 (Arbuckle, 2013) where the construct was developed (Jess, Totsika & Hastings, 2018).

Maternal parenting stress, related to having a child with a disability in the family was measured using the Parent and Family Problems sub-scale of the Questionnaire on Resources and Stress- Friedrich short form (QRS-F; Friedrich et al, 1983) (see Appendix VI). This subscale includes 20 items in total, coded as either true (0) or false (1). Five items were excluded as they have been identified as a robust measure of depression (Glidden & Floyd, 1997). This was to ensure that there was no overlap between the measures of stress and of mental health problems used in the present research. The QRS-F has good reliability when used with mothers of children with ID (Griffith et al., 2011). The Kuder-Richardson coefficient for the present sample was .86.

Maternal psychological distress was assessed using the Hospital Anxiety and Depression Scales (HADS; Zigmond and Snaith, 1983). This consists of two seven-item subscales that are rated from zero to three that measure levels of anxiety and depression. A unidimensional approach involves extracting a total score across all 14 items as a measure of psychological distress (Crawford et al, 2001). The HADS was initially developed to be used in outpatient settings but has been widely used in community-based research with parents of children with disabilities (e.g., Beck et al, 2004; MacDonald et al, 2010). The Cronbach's alpha for the psychological distress total score for the present sample was .88. For descriptive data at Time 1, see Table 2.1 and Table 2.2. For descriptive data at Time 2 see Table 3.2.

Table 3.2. Descriptive data for maternal outcomes at Time 2 in chapter 3

	HADS	QRS-F
Total	106	98
Mean Score (range; SD)	13.6 (1,31; 6.69)	5.05 (0-15; 3.84)

Data Analyses

Initially, I ran bivariate correlations between the main study variables (see Table 3.3). This was followed by a simple moderation analysis. PROCESS is a computational tool for path analysis-based moderation and mediation analysis. In this study, moderation analysis (model 1) was conducted using the PROCESS syntax (v2.16.3) developed for SPSS by Hayes (2013). Using this approach, moderation would be present if an interaction term between the putative moderator (maternal positivity) and risk factor (child behaviour problems) had a significant effect on maternal stress (parenting stress and psychological distress), thus potentially indicating a protective function of positivity. A significant main effect of positivity in the absence of a significant interaction term would indicate that positivity serves a compensatory function. The effect of maternal age, single parent status, employment status and maternal education (Time 1) on maternal stress was controlled for in the cross-sectional analysis. Maternal age, single parent status, employment status, maternal education and parenting stress/ psychological distress at Time 1 were controlled for in the longitudinal analysis. Child behaviour problems and maternal positivity were grand-mean centred prior to analysis to prevent multicollinearity. Bootstrapping (5000) was used to calculate standard errors and confidence intervals for all effects tested.

Results

I conducted two sets of analyses to address the research question and investigate the function of maternal positivity both cross-sectionally and longitudinally. Eight regression models were fitted in total, four cross-sectional and four longitudinal. Results of all analyses are summarised in tables 3.3 to 3.6.

Cross-sectional analyses

For the first set of analyses, four regression models were fitted (See Tables 3.3 and 3.4), varying the maternal outcome of focus (Time 1 parenting stress, or Time 1 psychological distress), and also varying the key child behaviour risk variable (BPI-01, or the Reiss Scales). All models were significant and the results of these analyses are summarised in Tables 3.3 and 3.4. Across all four regression models, maternal positivity only emerged as a significant moderator for one model: The interaction term between maternal positivity and frequency of child behaviour problems was significant when the outcome was parenting stress; $\beta = .008$, $t(111)= 2.69$, $p=.008$. Further output from the PROCESS syntax showed the relationship between child behaviour problems and parenting stress at high levels of positivity ($\beta= .15$, $t(111)=5.28$, $p=<.001$); mid-range levels ($\beta=.11$, $t(111)=5.35$, $p=<.001$); and low levels ($\beta=.06$, $t(111)= 2.83$, $p=.006$). This pattern was not as predicted by a protective function model: parenting stress in mothers with the highest levels of positivity was most strongly associated with the level of the child's behaviour problems.

Table 3.3. Associations (Pearson's correlation Coefficient) between child measures and maternal outcomes at Times 1 and 2.

	Maternal Positivity	Psychological Distress T1	Parenting Stress TI	Child Behaviour Problems	Child Mental Health	Psychological Distress T2	Parenting Stress T2
Maternal Positivity	1	-.58**	-.61**	-.35**	-.48**	-.47**	-.47**
Psychological Distress T1		1	.49**	.26**	.48**	.76**	.37**
Parenting Stress T1			1	.5**	.56**	.39**	.77**
Child Behaviour Problems				1	.77**	.23**	.48**
Child Mental Health					1	.35**	.51**
Psychological Distress T2						1	.49**
Parenting Stress T2							1

** *p= <.001*

Table 3.4. Regression analysis examining the cross-sectional compensatory and protective functions of maternal positivity between child mental health problems and maternal stress

Time 1 Predictor Variables	Time 1 Maternal Psychological Distress[a]				Time 1 Maternal Parenting Stress[b]			
	β	P	LLCI	UCLI	β	p	LLCI	UCLI
Child Age	.020	.885			-.307	.048		
Maternal Age	-.024	.765			.082	.377		
Single Parent Status	1.372	.014			.092	.883		
Maternal Education	-.652	.061			.343	.383		
Maternal Employment	.525	.580			-.285	.791		
Maternal Positivity	-.766	<.001	-1.028	-.505	-.899	<.001	-1.194	-.603
Child Mental Health	.098	.002	.038	.158	.188	<.001	.120	.256
Maternal Positivity x Child Mental Health	-.001	.826	-.014	.011	.013	.073	-.001	.027

LLCI= lower limit confidence interval, ULCI= upper limit confidence interval. [a] Model was significant: $F(8,118) =12.10$, $p=<.001$, $R2=.45$; [b] Model was significant: $F(8, 115) = 15.11$, $p=<.001$, $R2=.72$.

Table 3.5. Regression analysis examining the cross-sectional compensatory and protective functions of maternal positivity between child behaviour problems and maternal stress

Time 1 Predictor Variables	Time 1 Maternal Psychological Distress[a]				Time 1 Maternal Parenting Stress[b]			
	β	p	LLCI	UCLI	β	p	LLCI	UCLI
Child Age	.102	.489			-.232	.152		
Maternal Age	.087	.313			.067	.488		
Single Parent Status	1.259	.034			.086	.893		
Maternal Education	-.519	.163			.440	.282		
Maternal Employment	.345	.729			-.378	.731		
Maternal Positivity	-.937	<.001	-1.197	-.677	-.993	<.001	-1.278	-.708
Child Behaviour Problems	.019	.303	-.017	.056	.107	<.001	.067	.146
Maternal Positivity x Child Behaviour Problems	.005	.226	-.003	.012	.011	.008	.003	.019

LLCI= lower limit confidence interval, ULCI= upper limit confidence interval. [a] Model was significant: $F_{(8,114)}=9.60$, $p=<.001$, $R2=.40$; [b] Model was significant $(8,111)= 14.42$, $p=<.001$, $R2= .51$.

Table 3.6. Regression analysis examining the longitudinal compensatory and protective functions of maternal positivity between child mental health problems and maternal stress

Time 1 Predictor Variables	Time 2 Maternal Psychological Distress[a]				Time 2 Maternal Parenting Stress[b]			
	β	P	LLCI	ULCI	β	p	LLCI	ULCI
Child Age	-.041	.752			.163	.063		
Maternal Age	.087	.253			-.070	.169		
Single Parent Status	.398	.468			.488	.176		
Maternal Education	.184	.598			.333	.148		
Maternal Employment	-1.796	.041			-.055	.925		
Psychological Distress at Time 1	.792	<.001						
Parenting Stress at Time 1					.254	<.001		
Maternal Positivity	-.056	.695	-.339	.227	-.129	.180	-.320	.061
Child Mental Health	.030	.302	-.028	.089	.044	.039	.002	.085
Maternal Positivity x Child Mental Health	.003	.686	-.011	.016	-.001	.862	-.012	.010

LLCI= lower limit confidence interval, ULCI= upper limit confidence interval. [a] Model was significant: $F_{(9, 93)} = 18.14$, p=<.001, R^2=.64; [b] Model was significant: $F(9,84) = 11.62$, p=<.001, R^2=.56.

Table 3.7. Regression analysis examining the longitudinal compensatory and protective functions of maternal positivity between child behaviour problems and maternal stress

Time 1 Predictor Variables	Time 2 Maternal Psychological Distress[a]				Time 2 Maternal Parenting Stress[b]			
	β	p	LLCI	UCLI	β	p	LLCI	UCLI
Child Age	-.065	.612			.175	.050		
Maternal Age	.109	.155			-.078	.135		
Single Parent Status	.249	.644			.346	.338		
Maternal Education	.234	.496			.295	.212		
Maternal Employment	-.517	.080			-.064	.915		
Psychological Distress at Time 1	.777	<.001						
Parenting Stress at Time 1					.264	<.001		
Maternal Positivity	-.074	.603	-.356	.208	-.162	.099	-.355	.031
Child Behaviour Problems	.023	.183	-.011	.057	.022	.093	-.004	.048
Maternal Positivity x Child Behaviour Problems	<.001	.842	-.008	.009	-.004	.210	-.010	.002

LLCI= lower limit confidence interval, UCLI= upper limit confidence interval. [a] Model was significant: $F(9,90)=17.34$, $p=<.001$, $R^2=.63$; [b] Model was significant: $F(9,81)=11.89$, $p=<.001$, $R^2= .57$

Maternal positivity did have a significant negative effect on maternal psychological distress and parenting stress across all four regression models (see Tables 3.3 and 3.4). These main effect relationships between maternal positivity and maternal stress provide evidence that maternal positivity largely serves a compensatory function in these cross-sectional analyses. Thus, mothers who reported high levels of maternal positivity reported lower levels of psychological distress and parenting stress, controlling for the effects of child behaviour problems/mental health and several socioeconomic indicators. All child mental health and behaviour problems were significant predictors of maternal outcomes. Child mental health had a positive main effect on both parenting stress, $\beta = .19$, $t(115) = 5.49$, $p= <001$, and psychological distress $\beta = .10$, $t(118)=3.22$, $p=.002$, whilst frequency of child behaviour problems only had a significant positive main effect on parenting stress, $\beta =.11$, $t(111)= 5.35$, $p=<.001$.

Longitudinal analyses

Four regression models were fitted (See Tables 3.5 and 3.6) for the second set of analyses, varying the maternal outcome of focus (Time 2 parenting stress, or Time 2 psychological distress), and again varying child behaviour predictors. As with the first set of analyses, all regression models accounted for significant variance (see Tables 3.5 and 3.6). Across all four longitudinal regression models, maternal positivity did not moderate the relationship between child variables at Time 1 and maternal stress at Time 2. Thus, there was no evidence that maternal positivity functioned as a moderator longitudinally. Furthermore, maternal positivity did not have a significant main effect on later parenting stress or later psychological distress. Therefore, in this study, I found no evidence that maternal positivity served either a compensatory or protective function longitudinally. In addition, child mental health had a positive main effect on later parenting stress, $\beta = .04$, $t(84) = 2.10$, $p= .04$, but not on later psychological distress (see Table 3.6) whilst frequency of child behaviour problems did not

have a main effect relationship over time on either later parenting stress or later psychological distress.

Discussion

Research focusing on families raising children with ID has increasingly found that, despite challenges faced, positivity exists within these families. The present study addressed questions regarding the putative function of maternal positivity in mothers raising children with ID. I extended the original findings (Jess, Totsika & Hastings, 2018) by exploring the function of maternal positivity both cross-sectionally and longitudinally. Cross-sectional analysis found that maternal positivity had a direct association with maternal psychological distress and parenting stress. In addition, maternal positivity moderated the impact of child behaviour problems on maternal parenting stress. However, this interaction effect was not consistent with a putative protective function. The findings suggest that at a cross-sectional level, maternal positivity functions mainly as a compensatory factor. Thus, mothers who reported high levels of maternal positivity reported lower levels of psychological distress and parenting stress, controlling for the effects of child behaviour/mental health problems and several socioeconomic indicators. The findings are in concert with results from similar cross-sectional studies that demonstrate a main effect (compensatory) relationship (Lloyd & Hastings, 2008) using single indicators of positivity. For the first time, this compensatory relationship has been demonstrated using a robust latent measure of maternal positivity. I had also identified one moderation effect of positivity cross-sectionally using a latent measure of positivity. However, this moderated effect was not theoretically predicted and requires replication in future research.

The longitudinal analysis revealed different results. Maternal positivity did not have a direct effect on later maternal psychological distress or parenting stress and there was no evidence that positivity might function as a moderator over time. Although maternal

positivity had a compensatory function cross-sectionally, the direct effect of maternal positivity on maternal distress/ stress were small which potentially explains why a compensatory function was not evident in our longitudinal analysis.

I am unaware of existing studies within the family disability literature that have examined the protective function of positivity variables longitudinally for mothers or parents generally. In the small number of cross-sectional analyses published to date (Blacher & Baker, 2007; Weiss et al, 2015), single indicators of positivity were used. It is possible that different longitudinal results were identified primarily because of the use of a latent positivity construct. Findings from this study offer a valuable contribution to the wider understanding of maternal positivity and how it functions to potentially improve the well-being of mothers raising a child with ID. As discussed earlier, mothers of children with ID often report poorer well-being than mothers of typically developing children therefore it is of great importance that research continues to understand which constructs may improve well-being and indeed how. Future longitudinal research should examine the functions of both single indicators and latent positivity constructs to more fully understand the potential for a protective function of positivity. Importantly, the cross-sectional findings remained even after controlling for demographic characteristics (maternal age, child age, education, employment and single parent status) that previous research has suggested to be correlates of maternal mental health (Blacher et al, 1997; Elgar et al, 2007; Emerson & Llewellyn, 2001; Olsson & Hwang, 2001).

Within disability family literature there is overwhelming evidence to support the theory that child behaviour problems have an inverse relationship with maternal mental health both cross-sectionally and longitudinally. Although not the specific focus of this study, it is worth noting that this was partially true for the longitudinal analyses as child mental health had a significant effect on later parenting stress. The results of the cross-sectional

analyses were largely in concert with previous research confirming that child behaviour/mental health problems are correlates of maternal well-being.

Dealing with a child's behaviour problems is specific to the role of parenting whereas general psychological distress may be less affected by the challenges of raising a child with ID. This may explain why child behaviour problems had a significant effect on parenting stress but not on psychological distress. Similar results were found in a study of mothers of children with autism (Baker, Seltzer & Greenberg, 2011). Baker et al. found that whilst family adaptability, a similar construct to satisfaction with family, predicted a reduction of depressive symptomology, child behaviour problems did not have a significant effect on maternal depression.

In this study, child mental health problems had a negative main effect on both parenting stress and psychological distress cross-sectionally, and a longitudinal association with parenting stress. I was able to demonstrate that a construct of maternal positivity, generated from five single indicators of positivity which are both disability and non-disability specific, is associated with reduced present maternal well-being albeit not over time.

Limitations and Future Research Directions

A methodological limitation in the present study was that child behaviour problems and mental health measures were completed by mothers as were the measures of maternal well-being. Thus, this study suffered from a typical problem of shared source variance. The functions of positivity in future studies need to be explored in research designs where independent reports of child behaviour are obtained (e.g., from either the child's teacher or secondary caregiver). In addition, the sample size was modest. Therefore, the findings require replication before firm conclusions can be drawn about the functions of maternal positivity. There is a possibility that maternal positivity has only a small association with other

important variables in families of children with IDD potentially explaining why maternal positivity did not have a direct effect on maternal outcomes longitudinally. Dyches et al (2012) also found small to very small effects for the association between positive parenting and child outcomes in ID families. However, it is clear that positive constructs do require further study because they do not seem to simply represent the absence of negative outcomes. Similar conclusions have been drawn in other research where a differential pattern of associations for maternal emotional disorder and positive maternal mental health were found (Totsika et al, 2011b). Finally, the mothers were recruited via special schools supporting children with severe ID. The findings might be specific to this sub-group and samples covering the full range of ID should be included in future research.

I have argued that, given definitional and measurement issues, utilising a latent positivity construct is a methodological improvement. However, it is important to recognise an associated limitation that the application of a latent maternal positivity construct to different participant samples is problematic. The construct of maternal positivity is dependent on the participant sample. Therefore, replication of the findings is particularly crucial when developing latent positivity constructs in different samples and re-examining the functions of parental positivity. Furthermore, exploration of single indicators should continue to be investigated to understand how they function longitudinally.

Consideration also needs to be given to the time frame of this study. Although a longitudinal design, one year between data collection points may not be sufficient to detect a protective or compensatory function of maternal positivity if in fact it exists. Future research could benefit from longer time points or collecting data over multiple waves. Researchers would need to explore the function of positivity in larger and more representative samples as well as over longer periods of time.

Maternal positivity did have a significant negative effect on maternal psychological distress and parenting stress across all four regression models (see Tables 3.3 and 3.4). These main effect relationships between maternal positivity and maternal stress provide evidence that maternal positivity largely serves a compensatory function in these cross-sectional analyses. Thus, mothers who reported high levels of maternal positivity reported lower levels of psychological distress and parenting stress, controlling for the effects of child behaviour problems/mental health and several socioeconomic indicators. All child mental health and behaviour problems were significant predictors of maternal outcomes. Child mental health had a positive main effect on both parenting stress, $\beta = .19$, $t(115) = 5.49$, $p = <001$, and psychological distress $\beta = .10$, $t(118)=3.22$, $p=.002$, whilst frequency of child behaviour problems only had a significant positive main effect on parenting stress, $\beta =.11$, $t(111)= 5.35$, $p=<.001$.

Orientation to Chapter 4

The narrative surrounding raising a child with ID is fairly pessimistic. The literature suggests that caring for a child with ID is highly stressful and contributes towards poor parental mental health and family maladaptation (Dyson, 1997; Mitchell, Szczerepa, & Hauser-Cram, 2016; Patton et al, 2018). However, the previous two chapters were able to demonstrate that positivity does exist for mothers raising a child with ID and importantly that maternal positivity functions to improve maternal well-being. There have been consistent reports over several decades of relatively favourable outcomes for parents raising children with Down's syndrome than there are for other ID diagnoses. This is referred to as the 'Down's syndrome advantage'. Much of the literature pertaining to the Down's syndrome advantage focuses on negative outcomes such as parenting stress, depression and anxiety (Dabrowska & Pisula, 2010; Smith, Romski, Sevcik, Adamson & Barker, 2014). Very few studies have explored positive parental outcomes within the context of the Down's syndrome advantage.

Furthermore, there is emerging literature suggesting that the Down's syndrome advantage is the result of other variables as opposed to child diagnosis per se. There are very few studies which have examined whether the Down's syndrome advantage exists for positive outcomes in addition to negative ones. Chapters 2 and 3 evidence the presence of maternal positivity in spite of maternal stress. It was therefore considered important to investigate whether the Down's syndrome advantage existed for both positive and negative maternal outcomes when confounding variables were accounted for. Importantly, a group comparison design was utilised as advised by Hastings and Taunt's (2002) working model for further study of positivity. As discussed in chapter 1, Hastings and Taunt (2002) argued that identifying differences in positivity between groups is an important step to identifying how positivity functions. In addition, identifying differences is the initial step to take before investigating why these differences exist.

Chapter 4 utilised data collected during this PhD from families raising children in the UK with Intellectual Disability. Data were collected as part of a large UK based study called 'The 1000 Families Study.' Ethical approval was obtained by the NHS ethics committee (see Appendix XV) that was subject to annual reporting. This insured that the well-being, rights and dignity of participants were protected. Informed consent was obtained from all participants included in this study. All procedures followed were in accordance with the NHS Research Ethics Committee.

Chapter 4: The Down's syndrome Advantage and Positive Maternal Outcomes

Abstract

Background: Family members caring for children with Intellectual Disability report heightened levels of psychopathology, however those caring for children with Down's syndrome report comparably better outcomes. This is referred to as the Down's syndrome advantage. Very few studies have investigated whether the Down's syndrome advantage exists for positive maternal outcomes. This comparison design study examined whether the Down's syndrome advantage would be present for maternal psychological distress, closeness of parent-child relationship and perceived positive impact of child with ID when controlling for external variables.

Method: The sample consisted of mothers of children with Down's syndrome (*n*= 74) and mothers of children with an unknown ID diagnosis (*n*= 99) who completed measures pertaining to their own mental health, closeness to their child and perceived positive impact of their child on themselves and family unit (Positive Gains).

Results: A series of ANCOVAs revealed significant differences between mothers raising children with Down's syndrome and mothers raising children with an unknown diagnosis of ID indicating the presence of a Down's syndrome advantage in both negative and maternal outcomes. However, when child-related characteristics and external variables were controlled for, the Down's syndrome advantage only remained for positive gains.

Conclusions: It was concluded that the presence of diagnostic group differences in psychological distress and child-parent closeness were largely grounded in socioeconomic factors, child adaptive behaviours and child age. However, as the Down's syndrome advantage still remained for positive gains it is proposed that further investigation is required.

Mothers raising children with an Intellectual Disability (ID) experience higher levels of

stress, anxiety and more symptoms of depression than mothers raising typically developing

children (Baker et al, 2002; Eisenhower et al, 2005; Hayes & Watson, 2012; Totsika et al,

2011b). Such increased levels of maternal psychological distress begin early, from their

child's pre-school years and persist through to adulthood (Benson & Kersh, 2011; Estes et al,

2013; Orsmond et al, 2003). Within this group of mothers, there is some considerable

variation in the experience of psychological distress. One of the factors associated with

different patterns of maternal psychological distress is the nature of the child with ID's

genetic condition.

Evidence suggests that there is a clear differential impact of diagnosis on maternal

well-being. Such distinct patterns have been reported by Blacher and McIntyre (2006) who

identified differences in levels of maternal depression, negative impact and positive impact

between mothers of children with ID, cerebral palsy, Down's syndrome and autism. Similar

results were also reported in an earlier study conducted by Abbeduto and colleagues (2004),

and more recently by Griffith and colleagues (2010). These studies and others suggest that

mothers raising children with Down's syndrome (DS) have better outcomes than mothers

raising children with other conditions associated with intellectual disabilities (Blacher et al,

2013; Hodapp et al, 2003; Pisula, 2007). This pattern of better outcomes has commonly been

referred to as the 'Down's syndrome advantage' which has been evidenced in mothers across

their child's lifespan. (Dabrowska & Pisula, 2010). More recently there has been speculation

as to whether the Down's syndrome advantage is a robust diagnostic group difference or

whether it is driven by factors distinctly separate from the syndrome itself. A number of

factors could explain the apparent Down's syndrome advantage.

First, behaviour problems are often more frequent or severe in children with ID

(Baker et al, 2002; Totsika et al, 2011b; Totsika et al, 2014) and have long been associated

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with heightened maternal stress, anxiety and depression (Estes et al, 2009; Hastings, 2002; Johnston et al, 2003; Ricci & Hodapp, 2003; Tomanik, Harris & Hawkins, 2004). One potential explanation for the Down's syndrome advantage lies in the relatively fewer behaviour problems exhibited by individuals with Down's syndrome. Parents of children with Down's syndrome tend to report fewer child behaviour problems whilst simultaneously reporting better psychological well-being in their parents when compared to other ID groups. For example, Hodapp and colleagues (2003) reported that children with Down's syndrome had lower levels of behaviour problems compared to children with other ID diagnoses, and that mothers of children with Down's syndrome reported lower levels of child-related stress. Hodapp and colleagues (2003) concluded that child behaviour problems were strongly related to overall parenting stress. Such findings continue to be replicated, highlighting fewer behaviour problems in children and adults with Down's syndrome whilst simultaneously reporting better psychological well-being in their parents when compared to other groups of children with ID (Blacher & McIntyre, 2006; Glidden et al, 2014).

In addition to fewer behaviour problems, children with Down's syndrome often have comparatively higher levels of pro-social and adaptive behaviours which may in turn contribute to better maternal mental health (Beck et al, 2004; Blacher & McIntyre, 2006; Neece & Baker, 2008; Totsika et al., 2015). Using regression analyses, Blacher and McIntyre identified adaptive behaviours as accounting for differences in maternal negative impact between ID diagnostic groups demonstrating the significant association between adaptive behaviours and maternal well-being.

Family disability research has largely focussed on negative maternal outcomes, and the Down's syndrome advantage has predominantly been evidenced by lower levels of negative psychological outcomes in mothers. However, more recently researchers have examined the positive effects of raising a child with a disability (Corrice & Glidden, 2009;

Hastings, 2016; Hastings & Taunt, 2002; Ricci & Hodapp, 2003; Skotko et al, 2011). Positive outcomes have also demonstrated a putative Down's syndrome advantage in that mothers of children with Down's syndrome have reported that they are better rewarded by and have closer relationships with their child compared to other diagnostic groups (Hodapp, Ly, Fidler & Ricci, 2001). This was empirically evidenced by Abbeduto and colleagues (2004), who found that mothers of adolescents with Down's syndrome reported increased closeness with their child compared to mothers of adolescents with autism and Fragile X syndrome. Children with Down's syndrome are often described as sociable, cheerful (Walz & Benson, 2002) and affectionate (Wishart & Johnston, 1990), and it has been proposed that parents react favourably when their child with Down's syndrome displays a more 'Down's syndrome-like' personality (Hodapp, Ricci, Ly & Fidler, 2003). Thus, such positive characteristics may influence parents to be more affectionate towards their child, increase the amount of positive interactions they have with their child and encourage positive perceptions they have about their child. Arguably, the putative Down's syndrome advantage may be a direct consequence of increased perceived sociability in children with DS and thus would be evident in parents regardless of their child's diagnosis providing that the child exhibited higher levels of prosocial behaviours.

Despite the abundance of research demonstrating better mental health and greater positive outcomes for parents raising children with DS, caution should be taken before firm conclusions are made. There have been studies showing that once external factors are controlled, the DS advantage disappears. In a comparison study, Corrice and Glidden (2009) reported a Down's syndrome advantage in maternal well-being when compared with mothers of children with mixed ID aetiology However, group differences were no longer present when maternal age and child adaptive behaviours were controlled. In contrast, Eisenhower and colleagues (2005) found that mothers of pre-school children with DS reported less stress

and depression than mothers of other diagnostic groups (cerebral palsy and autism). When differences in behaviour problems were accounted for, child diagnoses still significantly contributed to maternal stress; providing evidence for the existence of a Down's syndrome advantage that could not be attributed to the child's behaviour problems. In the same study, Eisenhower and colleagues (2005) showed that ratings of positive impact were not significantly different amongst the groups of mothers in their study. These findings offer support for the putative Down's syndrome advantage in terms of negative outcomes (child behaviour problems, maternal stress) but not for a positive outcome measure (positive impact).

Much of the existing literature has been dominated by theories that have attributed a Down's syndrome advantage to the characteristics of the child. However, researchers have also examined other external factors. The majority of mothers of children with Down syndrome are more likely to have had their child at an older age (Loane et al, 2013). Notably older age in mothers of typically developing and disabled children have often been associated with better psychological adjustment (Benzies et al, 2013; Mayberry et al, 2007). In a recent study maternal age was found to be a significant predictor of positive family adjustment in that older mothers demonstrated higher levels of family adjustment after one year (Trute & Benzies, 2012). In accordance with this perspective, older mothers may have built up greater resilience and be better equipped to cope with the demands that come with raising a child with a disability, consequently demonstrating better outcomes than younger mothers. Thus, what is perceived as an advantage attributable to a diagnosis of Down's syndrome may actually be an advantage of maternal age.

Socioeconomic status could be considered as a factor associated with age. Older mothers and / or their partner may be more advanced in their career and have a comparatively better income. It is plausible that families raising a child with Down's syndrome are generally

of a higher socio-economic position than families of other ID diagnoses resulting in better maternal and family outcomes. In support of this hypothesis, Stoneman (2007) reported that household income was significantly higher for families raising children with Down's syndrome than in families raising children from other ID groups. Although not at the levels typically reaching statistical significance, other researchers have also identified families of children with Down's syndrome as having a higher family income than other ID households (Corrice & Glidden, 2009; Eisenhower et al, 2005; Glidden et al, 2014). Stoneman (2007) compared parental stress and depression of parents raising children with Down's syndrome to parents of children with ID of unknown aetiology. Results reflected a Down's syndrome advantage in that mothers of children with Down syndrome reported significantly lower levels of stress and depressive symptoms. However, this group difference disappeared when the variance attributable to family income was removed.

Contradictory findings emphasise the importance of accounting for demographic variables and child behaviours before firm conclusions are made about the existence of a Down's syndrome advantage. The aim of the current study was to determine whether the Down's syndrome advantage would be present in maternal outcomes when multiple child and maternal variables were controlled. Specifically, I compared mothers of children with Down's syndrome to mothers of children with ID of mixed unknown aetiologies. Much of the research that has identified a large Down's syndrome advantage has included an autism comparison group (Abbeduto et al, 2004; Griffith et al, 2010). This design is vulnerable to inflating the presence of an advantage. This is because autism is associated with significantly high rates of problem behaviours and impairments in pro-social skills and communication compared to children with ID. Therefore, I did not include children with autism in the comparison group for this study. In addition, I investigated the putative Down's syndrome advantage for both negative and positive maternal outcomes. I examined whether the Down'

syndrome advantage would be evident for maternal psychological distress, maternal positive gain and closeness in parent-child relationship when controlling for child adaptive and problem behaviours and family socioeconomic status. Furthermore, this study included a wide range of child ages and previously, younger child age has been associated with better maternal psychological adjustment (Goodman et al, 2011; Hodapp et al, 2003) therefore I also controlled for child age. I was unable to control for maternal age as I did not have access to this information. I hypothesised that any group differences found for maternal outcomes would be due to external factors. Considering the ongoing debate as to whether the Down's syndrome advantage occurs due to unaccounted external factors or child diagnosis, the primary question to be addressed in this study was, 'Does the Down's syndrome advantage exist when external factors have been accounted for?'

Method

Participants

Participants for this study were drawn from the Cerebra 1000 Families Study of families with a child with ID in the UK (see Appendix I). I selected a subsample of 173 mothers that identified themselves as either the biological, adoptive, or foster mother raising a child with ID of unknown aetiology or Down's syndrome. To be included in the comparison group, children with ID were selected if they did not have a named syndrome. Children with Down's syndrome were included if they also had a comorbid diagnosis of ASD. The total number of children ranged from 3 years to 15 years old (M=8.58 years, SD=3.03). The Down's syndrome group included 74 children of which 44 (59.5%) were boys and 30 (40.5%) were girls. Mean child age for this group was 8.21 years (SD=3.05). Most of the mothers raising a child with DS were married or living with a partner (85.2%) and either had a college or university education (86.5%). The ID group included 99 children of which 57 (57.6%) were boys and 42 (42.4%) were girls. Mean child age for this group was 8.85 years (SD=2.99).

Similarly, to the Down's syndrome group, the majority of mothers in the unknown aetiology group were married or living with a partner (81.8%) and educated to either college or university level (84.8%). See Table 4.1 for full demographic statistics.

Table 4.1. Mother and Child demographics

Participant Characteristics	Down's syndrome (*n*= 74)	Other Intellectual Disability (*n*=99)
Biological mothers	74 (100%)	89 (89.9%)
Adoptive mothers	-	9 (9.1%)
Foster mothers	-	1 (1%)
College or University qualifications	64 (86.5%)	84 (84.8)
Paid employment	42 (56.8%)	44 (44%)
Married or living with partner	63 (85.2%)	81 (81.8%)
Household income Median	6	6
Not managing financially	5 (6.8%)	14 (14.1%)
Could not raise £2000	14 (18.9%)	25 (25.3%)
SES composite Mean (range; SD)	*M*=.409 (-7 to 5; *SD*=2.63)	*M*=-.204 (-7 to 5; *SD*= 2.99)
Child age Mean (range; SD)	*M*=8.21 years (3-15; *SD*=3.05)	*M*= 8.85 years (3-15; *SD*=2.99)
Boys	44 (59.5%)	57 (57.6%)
Girls	30 (40.5%)	42 (42.4%)
DBC Mean (range; SD)	*M*=54.27 (16-109; SD=23.81)	*M*=68.67 (13-136; *SD*=29.63)
VABS communication score Mean (range; SD)	*M*=65.49 (34-108; *SD*=13.79)	*M*=60.58 (25-104; *SD*=14.34)
VABS socialisation score Mean (range; SD)	*M*=66.82 (38-101; *SD*=12.70)	*M*=59.27 (35-104; *SD*=11.58)

Note. Household income: Median of 6 in current study pertains to a weekly income of £600-700. At time of data collection UK gross weekly household income was £806 (Office of National Statistics). Managing financially: pertains to number of participants that indicated that they were either "Finding it quite difficult" or "Finding it very difficult." Raise £2000: pertains to participants that responded "I don't think I could raise the money" when asked how likely they would be able to raise £2000. SES is a composite variable created by incorporating 4 single item measures that captured; household income, if a family thought they could manage financially, financial hardship ("Raise £2000") and educational qualifications. Higher scores indicated better socioeconomic status.

Maternal measures

The Child-Parent Relationship Scale (CPRS; Pianta, 1992) (see Appendix VIII) is a 15-item measure completed by parents that assesses their perceptions of their relationship with their child. The items are rated on a 5-point scale and can be summed into two subscales that measure conflict and closeness of parent-child relationship. For this study, I used the closeness subscale. The 7-item closeness scale assesses the extent to which a parent feels that the relationship is characterised by warmth, affection, and open communication. Higher scores indicate greater levels of closeness. Cronbach's alpha for child-parent closeness in the current study was .82 for the DS group and .77 for the ID group.

The Positive Gain Scale (PGS; Pit-ten Cate 2003) (see Appendix IX) is a seven-item measure originally developed to assess positive aspects of raising a child with disability, on a five-point Likert scale. Five items reflect the perceived benefits of raising a child (e.g., "since having this child I have grown as a person"), and two reflect positive gains for the family (e.g., "since having this child, my family has become closer to one another"). Lower scores indicate greater positive gain. This measure has good reliability for mothers of children with ID (Macmullin et al, 2011). Cronbach's alpha in the current study was .85 for the DS group and .80 for the ID group.

The Kessler 6 (K6; Kessler at al., 2002) (see Appendix X) is a six-item measure developed to screen for the presence of psychological distress in non-clinical community samples. Participants were asked to score each item ranging from 0 (symptom not at all present) to 4 (symptom present over time) about psychological distress experienced in the past 30 days. Scores range from 0 to 24, with the higher scores indicating greater levels of distress. The K6 maintains excellent psychometric properties in mothers of children with ID (Totsika et al, 2011b) Cronbach's alpha in the current study was .79 for the DS group and .83 for the ID group. Please see Table 4.2 for descriptive data for outcome measures for mothers

of children with ID. Table 4.3 displays descriptive data for outcome variables for mothers raising children with DS.

Table 4.2. Descriptive data for outcome measures for other ID group

	CPRS	PGS	K6
Total	99	99	99
Mean Score (range; SD)	25.94 (7-35; 5.61)	13.47 (7-24; 4.07)	8.36 (0-22, 4.85)

Table 4.3 Descriptive data for outcome measures for DS group

	CPRS	PGS	K6
Total	71	74	74
Mean Score (range; SD)	28.34 (14-35; 4.88)	12.11 (7-27; 4.18)	6.35 (0-16; 4)

Socioeconomic Status (SES) (see Appendix XI) was measured using a composite variable created by incorporating four single item measures to capture multiple indicators of socioeconomic status. First, participants were asked to indicate in the survey how much their total weekly household income was with nine options starting from, 'Less than £200'; 'Between £200 and £300' and increasing in £100 increments to 'Over £1000'. The next single item asked participants to indicate how they thought they were financially managing on a 5-point Likert scale ranging from 'Living comfortably' to 'Finding it very difficult'. Higher scores indicated greater financial difficulty. A third item measured hardship. Participants were asked how likely they would be able to raise £2000 in one week on a 4-point Likert scale ranging from 'I could easily raise the money' to 'I don't think I could raise the money.' Higher scores indicated greater hardship. The final single item asked participants to report their highest educational qualification on a 6-point Likert scale ranging from 'No

qualifications' to 'University Degree'. Higher scores indicated higher qualifications. To create the composite variable, the items pertaining to financially managing and hardship were reversed scored. As all items were measured on different scales, they were all standardised-transformed, then summed to create the composite SES variable. Higher scores indicated higher SES.

Child measures

The Vineland Adaptive Behaviour Scale II- Survey form (VABS II; Sparrow et al. 2005) was used to measure child adaptive behaviour. This semi-structured interview measure contains a range of items that provide an assessment of adaptive behaviour across four domains: socialisation, communication, daily living skills and motor skills (used for children under seven years old only). These adaptive skills items are arranged in developmental order and not all questions are asked in an interview. 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. The socialisation and communication domain standard scores were used in the present analysis. Higher scores indicate greater adaptive behaviour.

The Developmental Behaviour Checklist- Parent (DBC-P; Einfeld & Tonge, 1992) is a 96 item measure designed to assess a broad range of behavioural and emotional problems in children and adolescents with ID (see Appendix XII). The DBC-P consists of five subscales; Disruptive/Anti-social Behaviour, Self-absorbed, Communication Disturbance, Anxiety and Social Relating. Each item is scored on a 3-point Likert scale ranging from 0 (not as far as you know) to 2 (very often or true). For the current study, I used the Total Behaviour Problem Score (TBPS) which is an overall measure of emotional and behavioural problems including all 96 items. The DBC-P has been shown to have good reliability in studies of children with ID (Einfeld & Tonge, 1992) and the Cronbach's Alpha for the current study was .73 for the DS group and .74 for the ID group.

Procedure

Study participants were recruited through multiple routes: special schools, social media advertising, and advertising via disability charities. Study packs distributed directly to parents (e.g., via the child's school) included an information sheet (see Appendix XIII), consent form (see Appendix XIV), the survey questionnaire (see Appendix I) and a prepaid return envelope. Participants could also request a pack to be sent to their home by following a link on social media. In addition, participants had the option to complete the survey online. Within the survey, participants were asked whether they would like to take part in a telephone interview and those who consented were contacted by a researcher to complete the VABS and DBC-P in the context of a semi-structured interview over the telephone. Due to the multiple methods used to distribute information about the Cerebra 1000 Families Study, no data are available on response rates.

Approach to Statistical Analysis

I conducted three sets of data analyses. The first was a bivariate Pearson Correlation analyses to check for multicollinearity between the predictor variables. I found no evidence of multicollinearity. Further analyses involved the comparison of the two ID groups: Mothers that had a child with Down's syndrome and mothers that had a child with ID of unknown aetiology. Independent sample *t*-tests were used to compare the mean scores of the three maternal outcome variables between the two ID groups. For the final set of analyses I conducted three analyses of covariance (ANCOVAs) that included all five covariates in each ANCOVA analyses to examine if any Down's syndrome advantage was robust to controlling for family and child variables.

Cohen's *d* was used to estimate the effect size of potential mean differences between the two ID groups. Cohen's *d* was estimated by calculating the mean difference between the

two ID groups, and then dividing the result by the pooled standard deviation. Confidence intervals for effect sizes were also calculated.

Results

Unadjusted Group Differences

Maternal psychological adjustment (positive gain, psychological distress, and close parent-child relationship) was compared between the two groups using *t*-tests to test for the presence of a putative Down's syndrome advantage. Mean scores for each group and Cohen's *d* effect sizes for the differences are summarised in Table 4.4. A statistically significant difference was present for all maternal outcomes. Mothers of children with Down's syndrome reported a closer relationship with their child; Cohen's *d* =.45, 95% CIs [.14, .76], more positive gain; Cohen's *d*= .33, 95% CI's [.03, .63], and less psychological distress; Cohen's *d* = .45, 95% CIs [.14, .75]. These unadjusted group comparisons support the hypothesis of a Down's syndrome advantage.

Table 4.4.

Means for maternal outcomes by ID group

Maternal Outcomes	Down's Syndrome		Other ID		Effect Size (*d*)	95% CI	
	Mean	SD	Mean	SD	Down's syndrome vs other ID	LL	UL
CPRS*	28.34	4.88	25.94	5.61	0.45	.14	.76
PGS*	12.11	4.18	13.47	4.07	0.33	.03	.63
K6*	6.35	4.00	8.35	4.85	0.45	.14	.75

*p =<.05. CI= confidence interval; LL = lower level; UL= upper level.

Adjusted Group Differences

After controlling for child behaviour problems, child communication and socialisation skills, family SES, and child age, group differences remained only for positive gain (see Table 4.5). Mothers of children with Down's syndrome reported more positive gain when compared with mothers of children with ID of unknown aetiology; Cohen's d= .37, 95% CIs [.05, .69]. None of the covariates included were significantly associated with maternal positive gain.

After controlling for child and maternal variables, there was no longer a main effect of ID group on child-parent closeness $F(1,143)$= 2.30 p=.132, Cohen's d= .24, 95% CIs [.09, .57] or maternal psychological distress $F(1,146)$=3.53, p=.062, Cohen's d= .32, 95% CIs [-.01, .64]. Increased child age $F(1,143)$=4.51, p= .035,) and child socialisation skills ($F(1,143)$= 16.06, p= <.001) were associated with higher levels of child-parent closeness. Lower composite SES scores were associated with increased maternal psychological distress $F(1,146)$=14.91, p= <.001).

Table 4.5. Analysis of Covariance Summary for all maternal Outcomes

Variables	CPRS closeness						Positive Gains						Psychological Distress					
	df	F	P	d	LLCI	ULCI	Df	F	P	d	LLCI	ULCI	df	F	p	d	LLCI	ULCI
Child age	1	4.51	.035				1	.783	.378				1	.931	.336			
DBC	1	.43	.515				1	.003	.953				1	.199	.657			
Communication	1	.86	.356				1	1.087	.299				1	2.449	.120			
Socialisation	1	16.06	<.001				1	1.544	.216				1	3.145	.078			
SES	1	.55	.46				1	.078	.781				1	14.914	<.001			
ID Group	1	2.29	.132	.24	.09	.57	1	4.866	.029	.37	.05	.69	1	3.533	.062	.32	-.01	.64

LLCI= lower level confidence interval; ULCI = upper level confidence interval; CPRS= child-parent relationship scale; DBC= developmental behaviour checklist; SES= socioeconomic status composite.

Discussion

The aim of this study was to explore further the putative Down's syndrome advantage, and in particular whether it is present in negative and positive psychological adjustment when I considered factors associated with but separate to Down's syndrome. A comparison design was adopted to explore the putative positive outcomes within the DS advantage framework. Hastings and Taunt (2002) proposed that to further a wider understand of positivity within families raising a child with ID, comparison designs must be adopted. Utilising this approach, unadjusted comparisons provided support for the existence of a Down's syndrome advantage for maternal positive gain, psychological distress, and perceived closeness of their relationship with their child. Thus, I found initial evidence of a Down's syndrome advantage for both positive and negative outcomes with effect sizes ranging from small to moderate. Subsequently, I examined whether the advantage would still be present once child and maternal factors were controlled. Findings indicated that effect sizes reduced and there was no longer a statistically significant difference between group means for either closeness of child-parent relationship and maternal psychological distress however differences remained for positive gain. By way of statistical significance, child age and socialisation skills explained some of the maternal group difference for parent-child closeness, and family SES explained some of the group difference for maternal psychological distress. However small effect sizes still remained for all maternal outcomes and increased for positive gains.

The results contrast with previous research that has found associations between fewer child behaviour problems and greater parent-child closeness (Abbeduto et al, 2004; Esbensen & Seltzer, 2011; Schuiringa et al, 2015) as well as lower maternal psychological distress (Estes et al, 2009; Harrison & Sofronoff, 2002). Although behavioural and emotional problems scores were lower for the Down's syndrome group (see Table 4.1), this variable was not associated with maternal outcomes once other factors were controlled. The results are

similar to the findings of Orsmond and colleagues (2006) who identified less social impairment as being predictive of a more positive mother-child relationship. The findings are also in concert with research that has demonstrated an association between child age and the quality of the mother-child relationship (Kim & Cicchetti, 2004). There is also some research to suggest that the parent-child relationship is at greater risk for problems at certain stages of the child's lifespan. In a longitudinal study of families with children who have autism, Taylor and Seltzer (2011) found the mother-child relationship improved over time whilst the child was in high school however became less positive when they left school. The availability of support services that are age-dependent may have had an effect on the child-mother relationship in the current study. Future research should consider the inclusion of support from services as a covariate to examine this possibility. The relationship between parent and child is likely to change throughout the child's lifespan thus it is unsurprising that child age is associated with closeness of child-mother relationship particularly as this sample included a wide range of child ages.

The findings in relation to SES, are similar to Stoneman's (2007) discussed earlier. Poorer maternal mental health in mothers of children with ID may be associated with socioeconomic status over and above child diagnosis differences (Emerson, Hatton, Llewllyn, Blacker & Graham, 2006; Olsson & Hwang, 2008). Although in the current study a composite SES variable was not associated with positive gain or relationship closeness, future research should explore other measures of SES or examine single indicators to identify whether there is a specific indicator of SES that has a significant effect on maternal well-being. It is likely that SES indicators included in the present study did not capture well socio-economic adversity. Low household income is not necessarily a robust indicator of income poverty as low levels of financial resources within a family may not necessarily place a family below the national poverty threshold.

Finding that the DS advantage remained for mothers' perceptions of positive gains may reflect a diagnostic group difference, particularly as differences between groups increased once covariates were controlled, emphasising further the effect of diagnostic group on maternal positive gains. Alternatively, the results may indicate that there are other covariates that were not included in this study which are important correlates of positive gain. For example, positive coping has been identified as a significant predictor of perceived positive gains in mothers of children with ID including mothers of children with Down's syndrome (Minnes et al, 2015).

Importantly, given that older mothers have been found to have better mental health outcomes than younger mothers maternal age may be a key factor in explaining what is perceived to be a Down's syndrome advantage and may go some way in accounting for the Down's syndrome advantage evident in the positive gain outcomes for this study. Other maternal factors, such as optimism, are related to positive psychological well-being in mothers of adults with Down's syndrome (Greenberg et al, 2004) and this should also be explored within context of a Down's syndrome advantage. Future research would benefit from including covariates such as coping, optimism and maternal age to determine whether the DS advantage is present for maternal positive gains or if that too is a product of factors separate to a diagnosis of Down's syndrome.

The DS advantage proposes that outcomes are better for families of children with DS than families raising children with any other ID diagnoses. Therefore, in this exploratory study the 'ID' comparator group included mixed ID diagnoses opposed to one specific ID diagnosis. However, in order to extend this study in future research, it would be an idea to explore whether the DS advantage still exists for positive gains when comparisons are made between homogeneous ID groups. Arguably covariates included in this study may interact with outcome measures differentially across ID groups. It is important that if these

differential interactions do exist that they are identified as they will have implications for clinical practice.

Folkman's theory of stress and coping proposes that during sustained periods of stress people search for positive meaning as a means of coping. With this in mind, future studies should investigate whether long periods of stress interacts with positive gains outcomes and whether that accounts for group differences. Although mothers of children with DS reported lower levels of psychological distress, stress was not measured or considered as a covariate or an outcome measure. Future studies may also investigate whether there are differences in reported positive gains scores between mothers experiencing long periods of stress and mothers that are not to explore potential differences and whether they are impacted by covariates.

In this study, I explored whether the DS advantage exists or could be explained by a variety of child and maternal factors. The findings further understanding about which factors may drive the presence of a Down's syndrome advantage.

Orientation to Chapter 5

The final study uses quantitative data from multiple sources. Having a methodological approach to measuring maternal positivity in chapter 2, its putative function in chapter 3 and whether single indicators of positivity were dependent on child diagnosis in chapter 4, a question remained about whether instruments designed to measure a construct and used in comparison studies worked equivalently across all groups.

Comparison design studies are particularly important because they offer valuable insight into group differences and help to attribute results to interventions (e.g., Chapter 4).

To determine whether an instrument is measuring the same construct it must be assessed for measurement invariance which indicates that the same construct is being measured across comparison groups. However, many researchers make the implicit assumption that an instrument is equivalent for all groups and therefore neglect this stage of analysis before utilising it in a study. Oversights such as these have important implications for conclusions based on findings taken from comparison-design studies. Given that in order to further our understanding of positivity within ID, comparison designs are essential, it is fundamentally important that the same construct is being measured across comparison groups to ensure valid conclusions.

With the growing interest in positive constructs within family disability research and the need for group comparison design research, chapter 5 assessed the measurement invariance of the Positive Gains Scale across three distinct populations: mothers of children with developmental delay, mothers of children with a chronic physical health problem and mothers of children without disabilities or health problems. This chapter further discusses the implications of measurement invariance testing for wider research.

Chapter 5 utilised in part, data collected during this PhD from families raising children in the UK with Intellectual Disability. Data were collected as part of a large UK based study called 'The 1000 Families Study.' Ethical approval was obtained by the NHS ethics committee (see Appendix XV) that was subject to annual reporting. This insured that the well-being, rights and dignity of participants were protected. Informed consent was obtained from all participants included in this study. All procedures followed were in accordance with the NHS Research Ethics Committee.

Secondary data were also utilised in chapter 5. Data were obtained with permission from Dr Ineke Pit-ten Cate's doctoral research. Further secondary data were obtained with the permission from Dr. Mike Petalas who had conducted a UK wide study of families that have a child with autism (Psychological adjustment and sibling relationships in siblings of children with autism spectrum disorders: Environmental stressors and the broad autism phenotype; Petalas, Hastings, Nash, Hall, Joannidi & Dowey; 2012). Applying for ethical approval to use the secondary data was not required however ethical considerations were taken into account. For example, all data were anonymised before being released to myself by the original researcher and the outcomes of the analysis did not allow for participants to be identified in any way.

Chapter 5: Testing for Measurement Invariance: Positive Gains Scale

Abstract

Background: Comparison studies offer vital insight into potential differences between groups. Due to such study designs there is now greater insight into differential levels of a construct between diagnostic groups. However, to accurately explore group differences it is imperative that an instrument is measuring the same construct between groups and that all groups interpret the construct in the same way. The present study investigated the measurement invariance of the 7-item Positive Gains Scale between three diagnostic groups.

Method: The sample consisted of three groups: mothers of children with ID (n=1148), mothers of children with a chronic physical health problem (n= 389) and mothers of children without disabilities or health problems (n=157). Testing for measurement invariance was conducted using Multigroup Confirmatory Factor Analyses (MGCFA) in AMOS 24.

Results: The factorial structure of the Positive Gains Scale was the same for all three groups. However, factor loadings of three items did not operate equivalently for all groups implying that it would be invalid to compare mean scores between

Conclusion: Findings indicated that the Positive Gains Scale works well to measure positive gains in single group studies. However, its utility in case-control studies is limited because although some of its items are comparable across groups, Positive Gains Scale total scores are not. It was concluded that testing for measurement invariance should be considered an essential preliminary investigation before any mean score comparisons are made.

One of the most common methodological paradigms of research that aims to describe the experiences of families raising a child with developmental disabilities (DD), is group comparisons to families raising a child without disabilities. Findings from such case-control studies suggested that mothers raising a child with DD experience higher levels of parenting stress and anxiety, in addition to more symptoms of depression than families raising a child without a disability (Hastings, 2016). In a large study with a UK-representative sample, (Totsika et al, 2011a) comparisons of maternal psychological well-being were made between mothers raising five year-old children with developmental disabilities (autism and intellectual disability) with mothers of children without disabilities. The researchers found that serious mental health illness and psychological distress affected a higher percentage of mothers raising children with DD than it did mothers raising children without disabilities. More recently, in a comparison study of parents raising 18 to 30 month old children with and without a developmental disability (Estes et al, 2013) it was found that parents raising toddlers with autism spectrum disorder (ASD) or DD reported more parenting stress than parents raising children without disabilities.

Although findings similar to those described above have been replicated throughout family disability literature (Eisenhower, Baker & Blacher, 2005; Hayes & Watson, 2011; Hoffman, Sweeney, Hodge, Lopez-Wagner & Looney, 2009) comparisons of maternal outcomes have also been made between a range of DD diagnoses groups to identify potential differences in maternal outcomes between diagnoses. For example, mothers of children with ASD have reported lower psychological well-being compared with mothers of children with cerebral palsy, Fragile X, and Down syndrome (Abbeduto et al, 2004; Griffith et al, 2010; Pisula, 2007).

In terms of explaining the variability in parental distress the evidence suggests that child behaviour problems are a significant factor when it comes to poor maternal well-being.

Baker and colleagues (2002) reported findings from mothers of children with and without a developmental disability. In that study, children with developmental disabilities were reported as having significantly more problem behaviours than the children without disabilities. Perhaps unsurprisingly, parenting stress was higher for parents raising a child with DD and this was largely accounted for by the presence of their child's behaviour problems and not their child's diagnosis. In another comparison study, Blacher and McIntyre (2006) investigated well-being in parents of young adults with developmental disabilities and found that although depression was lower for mothers of young adults with Downs syndrome than it was for other DD diagnoses (ID, cerebral palsy and autism), when behaviour problems were controlled for diagnostic group no longer accounted for the difference in maternal depression. Research continues to evidence the association between child behaviour problems and maternal mental health (McStay, Dissanayake, Scheeren, Koot & Begeer, 2014; Neece, Green & Baker, 2012).

Whilst findings from studies that focus on negative impact are useful in that they highlight challenges specific to DD families, they continue to perpetuate an unfavourable narrative and don't allow for a broader insight into the psychological well-being of these parents. Moving away from the focus on negative aspects associated with disability, there is a growing interest within disability family research on the positive aspects. In an earlier review of published positivity research at that time, Hastings and Taunt (2002) highlighted that although positive perceptions appeared to exist alongside negative experiences, positivity was largely a neglected area of research within the family disability field. The authors proposed a working model for further study of families' positive perceptions based on the suggestion that positive perceptions may function by moderating the impact of child disability on family members. Thus, it was suggested that to explore this function researchers must be able to measure positive perceptions and experiences explicitly. The review also highlighted that

case control designs could be useful for studying positive aspects of parental well-being across different populations.

Since the Hastings and Taunt (2002) review there has been an increasing effort to understand positivity in families of disabled children. To date, there have been a few controlled comparisons of parental positivity and results have been mixed. Mothers of children with DD have reported higher levels of positive impact than mothers raising children without disabilities (Blacher, Begum & Marcoulides, 2013). However, a number of studies tend to find no differences in levels of positive well-being between groups of mothers, despite differences in levels of mental health problems (Baker et al., 2002; Griffith et al, 2011; Totsika et al., 2011b).

As the understanding of these dimensions of maternal well-being increases, we need to intensify our efforts to define the positive aspects of raising a child with DD and to find appropriate tools for measuring these (Jess et al 2017). In existing case-control studies, comparisons often rest on the assumption that an instrument is measuring the same psychological construct in the same way across all study groups. However, this assumption is often not tested in practice.

The concept of measurement invariance suggests that a measure taps on to the same underlying construct across different groups of participants. This assumption is important for supporting the validity of group comparisons. Measurement invariance can be empirically tested. Evaluating measurement invariance assesses whether the dimensional structure of a construct (what an instrument is measuring) is the same for all groups, whether the factor loadings of the construct items are significant and whether the construct is manifested in the same way for all groups. Importantly, testing for measurement invariance assesses whether the response scale of an item is used in the same way for each group. If it is not, this suggests

that the construct does not have a common meaning and observed means cannot be compared amongst groups.

Measurement invariance requires a multistep process of assessments: configural invariance (tests that participants from each group conceptualise the construct in the same way), metric invariance (tests that the strength of the relationship between observed indicators and underlying construct is the same across groups), scalar invariance (needs to be established in order to compare means and indicates that participants that obtain the same score on a measure would obtain the same score on that measure's items irrespective of which group they belonged to) and error invariance (assesses whether the same level of measurement error is present for each item across groups).

Measurement invariance is an important assumption that needs to be tested before using instruments across different groups of participants in case-control designs. As an example, the Hospital Anxiety and Depression Scale (HADS: Zigmond & Snaith, 1983) is a fourteen item screening tool that is used to measure anxiety and depression in adults and in research investigating differences between mothers and fathers raising a child with DD (Hastings, 2003; Hastings et al, 2005a). The HADS has proven to be invariant across genders (Annunziata, Muzzati & Altoe, 2011; Hunt-Shanks, Blanchard, Reis, Fortier & Cappelli, 2010) indicating that it would operate equivalently for mothers and fathers when comparisons of parental psychological distress are made. However, it remains unknown as to whether the HADS is invariant for parents of children with and without disabilities. Similarly, the Strengths and Difficulties Questionnaire (SDQ: Goodman, 2001), which is frequently used to measure emotional and behavioural problems in children with DD (Emerson, Einfeld & Stancliffe, 2010; Kaptein, Jansen, Vogels & Reijneveld, 2008; Totsika, et al., 2011a) has been found to be invariant across race, gender, age and income groups (He, Burstein, Schmitz & Merikangas, 2013). However, like the HADS, it remains unknown whether the SDQ is

invariant across various DD diagnostic groups. As with instruments which measure negative

outcomes, it is unknown whether instruments that measure positivity are invariant for DD and

non-disability groups. Instruments that measure general, non-disability specific positivity

such as the Positive and Negative Affect Scale, the Scales of Psychological Well-Being and

Satisfaction with Life Scale have all been tested for invariance (Clench-Aas, Nes, Dalgard, &

Aarø, 2011; Crawford & Henry, 2004; Linley, Maltby, Wood, Osborne & Hurling, 2009)

with the latter found to be invariant across gender. However, like the HADS and SDQ it is

unknown whether any of these instruments measure the same construct in the same way for

developmentally disabled and non-developmentally disabled groups.

Research that has examined measurement invariance for research instruments used in

family disability research is very limited. To develop the research into the positive impacts of

raising a child with a DD, it is important that instruments are tested for measurement

invariance. The Positive Gains Scale (PGS; Pit-ten Cate, 2003) is one of the most frequently

utilised measures in family disability research to measure positivity (Griffith et al, 2011;

Jones, Totsika, Hastings, & Petalas, 2013; Weiss, MacMullin & Lunsky, 2015; Weiss &

Lunsky, 2011; MacDonald, Hastings & Fitzsimons, 2010). However, to date the

measurement invariance of the PGS has not been established. The aim of the current study

was to assess the measurement invariance of PGS across three distinct populations: mothers

of children with DD, mothers of children with a chronic physical health problem (CPHP) and

mothers of children without disabilities or health problems (termed thereafter typically

developing – TD). To examine the role of positive cognitions within both Resilience and

Stress and Coping frameworks it is important that an instrument that measures such

constructs works well and equivalently across groups so that valid comparisons can be made.

Therefore, the primary research question for this study was, 'Does the Positive Gains Scale

measure positivity equivalently for mothers of children with DD, mothers of children with a CPHP and mothers of TD children?'

Method

Participants

Participants in this study were mothers of children with DD, mothers of children with a CPHP and mothers of TD children. In total, 1694 mothers provided PGS data: 1148 (67.8%) were mothers of children with DD, 389 (22.9%) were mothers of children CPHP and there were 157 (9.3%) mothers of TD children (see Table 5.1 for participant information). In the TD group, there were 70 girls (44.6%) and 87 boys (55.4%) ranging from 5-12 years old with a mean age of 8.71 years ($SD= 1.81$). The CPHP group included 160 girls (41.1%) and 229 boys (58.9%) with an age range of 4-14 years and a mean age of 9.16 years ($SD= 2.34$). Forty-two (10.8%) children had spina bifida, 186 (47.8%) had hydrocephalus, 70 (18%) children were diagnosed with spina bifida and hydrocephalus, and 91 (23.4%) children had asthma. In the DD group there were 832 boys (72.5 %) and 316 girls (27.5 %) ranging from 2-17 years old with a mean age of 9.23 years ($SD= 3.1$). Seven hundred and nine (62%) children within the DD group had a diagnosis of Autism Spectrum Disorder (ASD). The remaining children included diagnoses of Down's syndrome, Cerebral Palsy, Global Developmental Delay and rare genetic syndromes including Fragile X.

Table 5.1 Participant Demographics

Participants	Developmental Disability	Physical Disability	Typically Developing
Total Mothers (*n*)	1148	389	157
Employed	506 (44.1%)	198 (50.9%)	129 (82.1%)
Unemployed	642 (55.9%)	185 (47.6%)	28 (17.8%)
No Qualifications	19 (1.7%)	87 (22.4%)	18 (11.5%)
Some GCSE'S	29 (2.5%)	-	-
5 or more GCSE'S	106 (9.2%)	120 (30.8%)	47 (29.9%)
GNVQ	-	34 (9%)	12 (7.6%)
A levels	131 (11.4%)	62 (15.9%)	31 (19.7%)
Higher than A level but below degree	222 (19.3%)	-	-
Degree	515 (44.9%)	62 (15.9%)	36 (22.9%)
Don't know	4 (0.3%)	-	-
Boys	832 (72.5%)	229 (58.9%)	87 (55.4%)
Girls	316 (27.5%)	160 (41.1%)	70 (44.6%)

Measures

The Positive Gains Scale (PGS; Pit-ten Cate 2003) (see Appendix IX) is a seven-item measure originally developed to assess positive aspects of raising a child with a disability based on the data from parents raising a child with spina bifida and/or hydrocephalus. Items are rated on a five-point scale. Five items reflect the perceived benefits for the individual parent (e.g., "since having this child I have grown as a person"), and two reflect positive gains for the family (e.g., "since having this child, my family has become closer to one another"). Lower scores indicate greater positive gain. Cronbach's alpha in the current study was .81 for the DD group, .80 for the CPHP group and .78 for the TD group.

Table 5.2 Descriptive data for the Positive Gains Scale for all 3 groups.

	ID Group	CPHP Group	TD Group
Total	1148	389	157
Mean Score (range; SD)	13.39 (7-34; 4.35)	15.86 (7-34; 4.75)	16.75 (7-32; 4.25)

Procedure

Participant data were extracted from multiple UK based studies. Mothers of the typically developing children were recruited from mainstream schools in England. Schools were asked to distribute questionnaires amongst parents of children aged 6-12. As some parents had more than one child attending the same school, the parent was asked to complete the questionnaire for their oldest child at that school.

Mothers of the children with a chronic physical health problem had completed a postal questionnaire previously as part of a comprehensive study concerning the developmental, behavioural and educational characteristics of children with these conditions

(see Pit-ten Cate & Stevenson, 1999). The initial sample was recruited through the register of

the Association for Spina Bifida and Hydrocephalus (ASBAH). Families were entered on this

register when they contacted ASBAH for information and/or support. The current sample

included mothers who indicated they would be interested in taking part in future research.

Mothers were also recruited via the asthma clinic at a large General Hospital. Families were

identified using in- and outpatient record sheets. Surveys were sent to families with a child

with asthma aged 5-13 years. Initially only families of 6-12 year old children were contacted,

however, as only a relative small number of questionnaires was returned, the age range was

extended to also include families of children aged 4 and 14 years and 173 (45%) were

reported as having a learning problem.

In the DD sample, 947 mothers were drawn from the Cerebra 1,000 Families Study,

which is a UK-wide survey of families with a child with intellectual disability. An additional

201 mothers were from a UK-wide study of families who have a child with autism (Petalas et

al, 2012). Mothers of the children with DD were recruited through multiple routes: special

schools, social media advertising, and advertising via disability charities. Survey packs

distributed directly to parents (e.g., via the child's school) included an information sheet,

consent form, the survey questionnaire and a prepaid return envelope. Participants could also

request a survey pack to be sent to their home. In addition, participants had the option to

complete the survey online.

Approach to statistical analyses

All analyses were conducted in AMOS 24 (Arbuckle, 2016). Testing for the measurement

invariance of the PGS across all three groups involved a multi-step process in which equality

constraints were increasingly imposed at each stage as recommended by Byrne (2009).

To evaluate the dimensional structure of the PGS (see figure 5.1), I performed confirmatory factor analysis (CFA) using the data from each of the three groups separately thus creating a baseline model. Results indicated a reasonable model fit of the data within each group separately. However, I believed I could improve the model fit for each group by reviewing the CFA specification: I examined bivariate correlations between PGS items in each group separately to check whether any PGS items were significantly correlated with one another. Family-level gains items 4 ("Since having this child, my family has become closer to one another") and 5 ("Since having this child, my family has become more tolerant and accepting") correlated strongly ($r = \geq .5$) within each group, therefore their error terms were allowed to correlate in the baseline models (see Figure 5.1). Although correlating error terms should be avoided (Gerbing & Anderson, 1984) as they produce multidimensional factor scores that can be difficult to interpret, in this case the content similarity of Items 4 and 5 indicated that they represented an alternative to a very similar theme. CFAs with the correlated error terms demonstrated an improved fit (see Table 5.3). The final step involved fitting the last model to a randomly selected subgroup within each group separately to ensure that the results could be replicated. Results from the random sample CFA indicated a good fit with the DD data ($\chi^2(13) = 24.96$; CFI=.97; RMSEA= .070, 90% CI [.025, 112]), a good fit with the typically developing data ($\chi2(13)=13.13$; CFI=.99; RMSEA=.010, 90% CI [<.001, .095]) and a good fit with the chronic physical health problem data, ($\chi2(13)=17.37$; CFI=.99; RMSEA=.041, 90% CI [<.001, .086]). As fit indices demonstrated a good fit, this indicated that the structure of the PGS was supported in all three groups.

Figure 5.1. Baseline model of the Positive Gains Scale
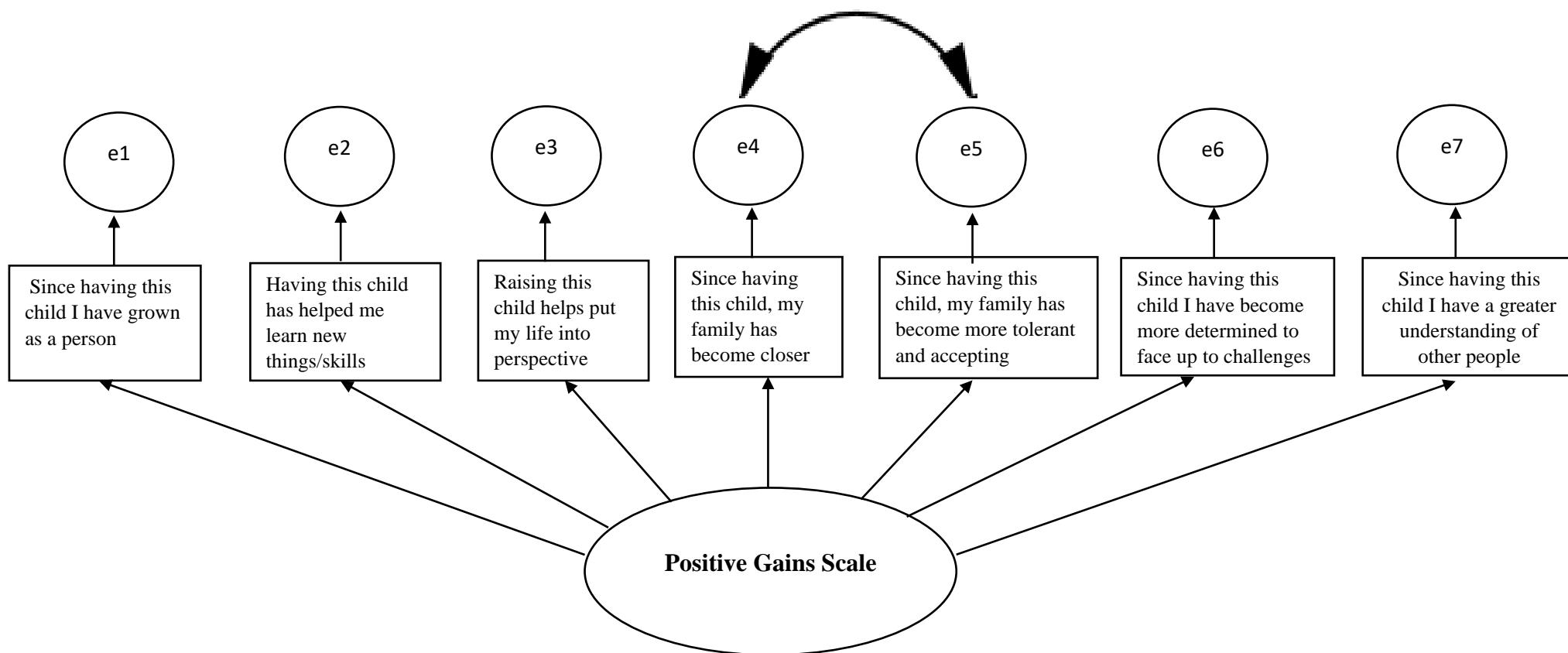
Table 5.3. Summary of goodness-of-fit statistics in determination of Baseline Models

Model	χ2	df	CFI	RMSEA	90% CI	ECVI
Intellectual Disability						
Unconstrained Model	427.156	14	.847	.160	.148, .174	.409
Error terms for Items 4 and 5 correlated	86.809	13	.973	.070	.057, .085	.114
Physical Disability						
Unconstrained Model	50.405	14	.944	.082	.058, .107	.238
Error terms for Items 4 and 5 correlated	19.021	13	.991	.035	.000, .066	.162
Typically Developing						
Unconstrained Model	31.414	14	.926	.089	.047, .131	.471
Error terms for Items 4 and 5 correlated	20.982	13	.966	.063	.000, .110	.417

df = degrees of freedom. CFI= comparative fit index. RMSEA= root mean square error approximation. CI= confidence intervals, ECVI= expected cross-validation index. Item 4= "Since having this child, my family has become closer to one another", Item 5= "Since having this child, my family has become more tolerant and accepting".

Testing for measurement invariance was conducted using Multigroup Confirmatory Factor Analyses (MGCFA) in AMOS 24. This method involves constraints being imposed successively on the model at each stage of analysis: Configural invariance (tests that participants from each group conceptualise the construct in the same way and is the model to which subsequent models are compared), metric invariance (tests that the strength of the relationship between observed indicators and the underlying construct is the same in each group), scalar invariance (tests that the response scale is used in the same way by participants across groups; it needs to be established to compare means across groups. Scalar invariance would indicate that participants that obtain the same score on a measure would obtain the same score on that measure's items irrespective of which group they belonged to), and error invariance (assesses whether the same level of measurement error is present across all groups).

Configural invariance assumes that the structure of the PGS is equal across groups. To test for configural invariance, the baseline model was fitted to data from all three groups simultaneously. This model is tested by constraining the factorial structure to be the same across all three groups. The configural model provided one set of fit statistics for the overall model to which subsequent models were compared for difference of fit. In large samples, the risk of a Type I error is present if conclusions are made on the basis of the $\chi 2$ test only (Hoyle & Panter, 1995). Therefore, the criteria for testing whether the assumption of configural invariance holds were a non-significant $\Delta\chi2$ and a ΔCFI equal to or less than 0.01, as suggested by Cheung and Rensvold (2002). Delta (Δ) indicates the test statistic tests a model difference.

Metric invariance assumes configural invariance and equality of factor loadings. Thus, metric invariance suggests that the relationship between observed variables and the

underlying construct of positive gains which is operationalised by the factor loading is similar across groups. To test this hypothesis, equality constraints were imposed on the factor loadings in all groups. Lastly, I tested for scalar invariance which assumes metric invariance and equality of intercepts across all groups. Equality of intercepts indicates that if participants from different groups obtained the same PGS score, all three groups use the response scale of the indicators in the same way. Therefore, scalar invariance suggests that the PGS has a common meaning across the three groups. Support for scalar invariance indicates that PGS means can be meaningfully compared among groups.

If full invariance could not be demonstrated, I examined whether partial invariance was possible, as proposed by Byrne, Shavelson and Muthen (1989). Partial invariance tries to identify which of the factor loadings or intercepts are different across groups. Byrne and colleagues argued that full metric invariance is not necessary to continue further tests of invariance providing that at least one item is metrically invariant. If partial metric invariance is achieved, partial scalar invariance can be tested for. It was further proposed that if there were at least two factor loadings and intercepts constrained equal across groups, valid inferences regarding group mean differences can be made (Byrne et al, 1989).

To evaluate model fit I used several criteria: a Root Mean Square Error of Approximation (RMSEA) under .08 (Browne & Cudeck, 1993), a Comparative Fit Index (CFI) above .95 (Hu & Bentler, 1999) and an Expected Cross-validation Index (ECVI) as recommended by Byrne (2009).

Results

Configural Invariance Testing

Once a baseline model was established, I then moved to multigroup CFA (MGCFA) to cross-validate the baseline model structure across the three groups simultaneously. Results indicated a good fit with the data ($\chi^2(39) = 125.231$; CFI =.98; RMSEA=.036, 90% CI [.029, .043]. Thus, configural invariance was achieved indicating that the factorial structure of the PGS was the same for all three groups.

Metric Invariance Testing

When equality constraints were imposed on all factor loadings the $\Delta\chi2$ was significant and the ΔCFI was greater than .01. Thus, the imposition of constraints resulted in statistically significant decreases in the model fit when compared to the configural model.

Significant differences were identified between groups in factor loadings associated with Items 3 ("Raising this child helps put my life into perspective"), 5 ("Since having this child, my family has become more tolerant and accepting") and 7 ("Since having this child I have a greater understanding of other people").

As recommended by Van de Schoot, Lugtig and Hox (2012), I released constraints on factor loadings with the largest unstandardized differences and continued to release subsequent factor loadings until I identified the items which caused measurement invariance not to hold. A non-significant $\chi2$ difference ($\Delta\chi2$ (8) =13.92; p=.08) and a ΔCFI <.01 was achieved when constraints were released on factor loadings for Items 3 ("Raising this child helps put my life into perspective") and 7 ("Since having this child I have a greater understanding of other people") across all groups. Thus, partial metric invariance was

achieved indicating that equivalence of factor loadings is present across all three groups

except for Items 3 and 7.

Table 5.4. Goodness-of-fit statistics for Tests of Metric Invariance

Model	χ2	Df	CFI	RMSEA	90% CI	Δ χ2	ΔCFI
A1 (all factor loadings held equal across groups)	190.548	51	.961	.040	.034, .046	65.317*	.015
B1 (equality constraints released on factor loadings of Items 5&7)	150.520	47	.971	.036	.030, .043	25.289*	.005
C1 (equality constraints released on factor loadings of Items 3 & 5)	153.159	47	.970	.037	.030, .043	27.928*	.006
D1 (equality constraints released on factor loadings of Items 3 & 7)	139.147	47	.974	.034	.028, .041	13.916	.002

*$p<.001$. df = degrees of freedom. CFI= comparative fit index. RMSEA= root mean square error approximation. CI= confidence intervals.

Item 3= "Raising this child helps put my life into perspective", Item 5 = "Since having this child, my family has become more tolerant and accepting", Item 7= "Since having this child I have a greater understanding of other people."

Scalar Invariance Testing

The partially invariant model (constraints released on factor loadings for Items 3 and 7) observed in the previous step was carried forward as the default model to test for partial scalar invariance. Initially, constraints were imposed on all intercepts except for Items 3 and 7. Comparison of model fit revealed a significant χ^2 difference ($\Delta\chi2$ (22) = 540.86; p=<.001) and a ΔCFI greater than .01 (ΔCFI= .15), suggesting that the fit was significantly worse than that of the configural model. Subsequently, each intercept was examined for group invariance. There were significant $\chi2$ differences and a ΔCFI greater than .01 for all combinations of intercept constraints. Therefore, partial scalar invariance was not achieved indicating that PGS mean scores cannot be meaningfully compared between groups. As scalar invariance was not achieved, testing for error invariance could not be conducted.

Discussion

In recent years, there has been a growing interest in positivity associated with raising a child with disabilities. To further understand the construct and dimensions of positivity case-control studies are needed and within those researchers need to have access to instruments that operate equivalently across different study groups. In this study, I explored measurement invariance of the PGS, a scale that measures parents' perceptions of positive gains they experience as individuals or families since having their child. The aim was to determine whether the PGS operated equivalently for mothers of children with DD, mothers of children with a CPHP and mothers of children without any health problems or disabilities.

In the first step of investigation I demonstrated that the PGS construct had a good fit with the data from each group of participants, especially when items 4 ("Since having this child, my family has become closer to one another") and 5 ("Since having this child, my

family has become more tolerant and accepting") were allowed to co-vary; both these related to family-level positive gains.

This study then assessed the measurement invariance of the PGS using Multigroup Confirmatory Factor Analysis (MGCFA). The findings supported configural invariance indicating that the factorial structure of the PGS was equal across groups. However, factor loadings of three items (Items 3, 5 and 7) appeared not to be operating equivalently for all groups, which meant that I was not able to demonstrate full metric invariance across groups. However, when constraints were released for factor loadings of Items 3 and 7, partial metric invariance was achieved and Item 5 operated equivalently across groups. Partial scalar invariance could not be demonstrated, indicating that PGS total scores cannot be meaningfully compared between mothers of children with DD, mothers of children with a CPHP and mothers of children without health problems or disabilities.

Although initially, it appeared that mothers of typically developing children were less positive about their child than mothers of children with ID (see Table 5.2), given study findings, comparisons such as these are invalid and offer no insight into PGS scores relative to comparator groups. However, the findings do suggest that the PGS works well to measure positive gains in single group studies. Utilising the PGS in case-control studies is limited because although some of its items are comparable across groups, PGS total scores are not. Importantly, despite the fact that PGS mean scores cannot be compared across groups in this instance, comparing scores of equivalent items would still offer useful insight into potential group differences and could be adopted as an alternative when mean score comparisons should not be made.

Although this study focused on measurement invariance of the PGS, the findings have wider implications pertaining to the validity of case-control studies. As earlier discussed,

many case-control studies within the disability literature utilise instruments that measure parental well-being making comparisons between those raising children with and without disabilities (Eisenhower, Baker & Blacher, 2003; Estes et al, 2013; Totsika, Hastings, Emerson, Berridge & Lancaster, 2011). However, these comparisons are often made in the absence of a definitive answer as to whether that instrument is indeed invariant across these groups. The findings suggest that future research should examine the invariance of any measure implemented in a comparison study to ensure valid comparisons are being made. Furthermore, as suggested in the previous chapter, if future studies are going to investigate Folkman's theory of stress and coping further by adopting a group comparison paradigm then it is fundamentally important that an instrument that measures positivity, such as the PGS, works in the same way for all groups to ensure that findings are valid. Limitations of this study should also be considered when interpreting the results. The development of the PGS was based on data taken from families of children with a chronic physical health problem. It could be argued that it is more appropriate to assess the Positive Gain Scale's measurement invariance within clinical samples only and in this study, between different disability types such as physical and developmental rather than including a typically developing group. The underlying concept of positivity may in fact be very different for parents raising a child without health problems and more similar for parents raising a child with health problems or a disability. In addition, the concept of positivity may also be different between the ranges of DD diagnoses included in this study.

The outcome of partial metric invariance suggests that the strength of the relationship between Items 3 ("Raising this child helps put my life into perspective.") and 7 ("Since having this child I have a greater understanding of other people.") and the underlying construct of positive gains is different for each group, indicating that the PGS is not manifested in exactly the same way in each group and it is important to understand why.

Partial metric invariance may have occurred due to participant's different response styles which can affect response variability (Liu, Harbaugh, Harring & Hancock, 2017). Extreme response style (ERS) refers to a tendency for participants to select the extreme ends of a scale, (i.e., strongly agree, strongly disagree) and non-extreme response style (NERS) which occurs when participants avoid selecting the extreme ends of a scale (i.e., neither agree nor disagree). Arguably parents raising a child with a disability are more inclined to report on the extreme ends of the scale for Items 3 and 7 because these statements are more relevant to this group of mothers whereas mothers raising typically developing children may have adopted a NERS. These points raised may lend an explanation as to why in this study scalar invariance could not be attained for all three groups. This is something to explore in future research.

Lastly, participants in this study were recruited from multiple sources and the limitations of this approach should be addressed. Online recruitment proved to be the most successful method thus contributing to the overrepresentation of mothers raising children with ID compared to the other diagnostic groups (see Table 5.1). Considering this, a targeted online approach to recruiting mothers of TD children and mothers of children with CPHP may have yielded higher participant numbers across groups and should be considered for future research. Furthermore, mothers of children with asthma may have responded differently to the PGS dependent on whether their child was an inpatient or an outpatient if the presumption is that inpatients have the most severe cases of asthma. This aspect of study design is also worthy of further exploration as it could have had an impact on reported findings.

To my knowledge, this is the first study that has investigated measurement invariance in a disability-specific positive instrument. Further examination into the psychometric

properties of similar instruments is important to enable researchers to measure positivity effectively and to make valid group comparisons.

In conclusion, although the Positive Gains Scale has strong psychometric properties as evidenced by the consistently high Cronbach's alpha achieved across a range of populations (Griffith et al, 2011; Jones, Totsika, Hastings & Petalas, 2013; Minnes, Perry & Weiss, 2015; Weiss, MacMullin & Lunsky, 2015), future studies should remain cautious if mean scores are compared between groups and measurement invariance has not been determined, not just for the Positive Gains Scale but all measures implemented within a study. Indeed, comparison studies within family disability research are needed as they provide important insight into potential differences and similarities which are imperative to a wider understanding of families raising a child with DD. Therefore, testing for measurement invariance should be considered an essential preliminary investigation before any mean score comparisons are made.

Chapter 6: Overall Discussion

Overview

Raising a child with intellectual disability involves a unique set of challenges and difficulties which can often contribute to greater symptoms of parental psychological distress when compared to parents raising a child without disabilities (Eisenhower, Baker & Blacher, 2005; Estes et al, 2013; Olsson & Hwang, 2001). However, in the face of this many parents report positive feelings about their child, stating that their child has had a positive effect on both themselves and their family (Hastings, 2016; Stainton & Besser, 1998). Folkman's theory of stress and coping offers an explanation for these feelings of positivity in the face of sustained stress whilst resilience theory explains how positivity can be utilised to support maternal well-being. As mentioned throughout this thesis, it is only in recent years that researchers have explored positivity within families raising a child with disabilities, particularly positivity specific to mothers. The fundamental purpose of this thesis was to address some of the gaps in the relatively new field of maternal positivity within the context of intellectual disability.

With the theoretical framework of Resilience in mind, this thesis begun to explore the concept of maternal positivity, what that might look like for mothers raising a child with intellectual disability and how it potentially functions. Although the function of single indicators of positivity have been explored in recent years, this thesis demonstrates for the first time that a latent construct of maternal positivity may have value in research. This thesis further explored positive maternal outcomes within the context of the Down's syndrome advantage, which proposes that parents raising a child with DS have better outcomes than parents raising children with other ID diagnoses. Again, there are very few studies which have researched positivity in this context. With the growing interest and awareness of maternal positivity further research must continue. Importantly, to understand how positive constructs function within the context of resilience and/ or stress and coping frameworks, instruments that measure such constructs must work equivalently across comparison groups.

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Thus, the final empirical study of this thesis explored the psychometric properties of an increasingly popular instrument, The Positive Gains Scale, designed to measure the positive perceptions of caregivers raising a child with disabilities. This thesis has attempted to expand existing knowledge of positivity specific to mothers raising a child with ID and address some of the unanswered questions in the literature.

Summary of research findings

Chapter 2 tested whether positivity can be measured as an overall construct that is made up of several indicators that span across subjective well-being to parenting role perceptions. Despite including an indicator that exclusively measures positivity specific to a child with disabilities (positive contributions), the indicator that had the strongest association with the latent factor was parental self-efficacy. Therefore, maternal perceptions of how they parent their child may be particularly important to their overall feelings of positivity, more so than positive perceptions they have specifically about their child with ID. The results also showed significant associations between the latent construct of maternal positivity and child and maternal outcomes. Negative associations between maternal positivity and maternal psychological distress and child behaviour problems were found in addition to a positive association between maternal positivity and child pro-social behaviour. The direction of associations evidenced in this study followed that of research using single indicators of positivity. As causality could not be inferred from the findings, a need for a longitudinal design study was identified to further understand the relationship between maternal positivity and child behaviours and importantly for this thesis, the relationship between maternal positivity and maternal psychological well-being.

Chapter 3 further explored the underlying latent construct of maternal positivity by investigating its function within the context of resilience theory both cross-sectionally and

longitudinally. Maternal positivity was explored as a potential compensatory and/or a protective factor against child behaviour problems which were framed as a risk factor for maternal psychological distress and parenting stress. This chapter evidenced support for the argument that cross-sectionally, maternal positivity has a largely compensatory function. Findings were that maternal positivity had a direct negative effect on maternal psychological distress and parenting stress, demonstrating a compensatory function. However maternal positivity was also identified as having a protective function as it moderated the impact of child behaviour problems on parenting stress cross-sectionally. These findings were in line with similar cross-sectional studies that have used single indicators of positivity (Ekas, Lickenbrock & Whitman, 2010; Kayfitz, Gragg and Orr 2010; Lloyd & Hastings, 2008). Furthermore, findings from this study contributed to the wider body of family disability literature, drawing on the previous study, evidencing that maternal positivity exits independently of poor mental health outcomes and that it can function to alleviate the adverse effects of child behaviour problems on maternal mental health. Results from the longitudinal analysis did not support all of the cross-sectional findings in that maternal positivity did not function as either a compensatory or protective variable over time. This study was unique in that it was the first study to examine the functions of maternal positivity using a multi-indicator latent positivity variable.

In chapter 4 the focus moved from the function of positivity to exploring the potential differences in the levels of maternal positivity between diagnostic groups. Research on the Down's syndrome advantage represents a history of positively orientated studies examining whether parents raising a child with Down's syndrome have better outcomes than parents raising children with other ID diagnoses. Reduced depression and fewer symptoms of anxiety have been reported by parents raising children with Down's syndrome compared to parents of children with ASD for example (Abbeduto et al, 2004; Griffith et al, 2010). There is,

however, an emerging argument within the literature that the presence of a Down's syndrome advantage is a consequence of external factors such as child and maternal characteristics and not perhaps directly related to the child's diagnosis. A majority of previous studies have identified the Down's syndrome advantage with a focus on negative parental outcomes such as parental stress and anxiety. Chapter 4 was unique in that it investigated the Down's syndrome advantage in positive <u>and</u> negative parental outcomes. The aim of this study was to determine whether the DS advantage was evident when external factors were controlled for and whether it would be evident in positive maternal outcomes. It was found that the Down's syndrome advantage was indeed apparent when external factors were not controlled for in that mothers of children with Down's syndrome reported a closer relationship with their child, more positive gain, and less psychological distress than mothers of children with other intellectual disabilities. However, when child behaviour problems, child communication and socialisation skills, family socio-economic status (SES), and child age were controlled for the DS advantage was no longer present for child-parent closeness or maternal psychological distress. Increased child age and socialisation skills accounted for higher levels of child-parent closeness whilst lower SES accounted for group differences in psychological distress. Even when child and maternal characteristics were controlled for, mothers of children with DS perceived their child more positively than mother of children with other intellectual disabilities. The importance of this study is that the findings contribute to a changing narrative surrounding parenting a child with ID. Whilst raising a child with ID has largely been promoted as a negative experience in which parents of DS children are 'better off,' this chapter was able to demonstrate that differences in parental outcomes are related to external factors and not necessarily the child's diagnosis. However, this study did provide support for the DS advantage in relation to maternal positive perceptions of their child.

Chapter 5 was the final empirical study of this thesis which looked at the measurement invariance of the Positive Gains Scale (PGS: Pit-ten Cate, 2003). It was found that the structure of the PGS looked the same for mothers of children with ID, mothers of children with a physical health problem and for mothers of children without disabilities when two of the family items were allowed to correlate. Upon further analysis, not all PGS items operated equivalently for all groups, thus demonstrating partial metric invariance. However partial scalar invariance was not achieved which meant that group means could not be compared as the underlying construct of the PGS does not have the same meaning for all groups. Thus, this chapter was of great importance as it highlighted potential flaws not only in group comparisons that use the PGS but for wider group comparison design studies that have utilised instruments not assessed for measurement invariance.

Theoretical and methodological implications

Within the existing ID family literature there is no clear consensus on what maternal positivity is, how it is defined, how it is measured and how it functions. One aim of this thesis was to explore an empirical method through which 'positivity' could be approached quantitively. Previous research has attempted to define positivity as either an absence of negativity or only explored a specific focus of positivity such as family cohesion or positive perceptions of the child. Chapter 2 concluded that maternal positivity can be measured as a latent construct (in this case consisting of five single indicators of positivity that focus on three specific domains of maternal life, parenting, family and general well-being).Chapter 2 showed that when mothers of children with ID feel efficacious about their parenting, are highly satisfied with their own and family life, have a general positive mood and have positive perceptions of their child, that they will experience greater 'maternal positivity'. Therefore, contrary to maternal positivity simply being the absence of negativity, this thesis posits that maternal positivity is a complex and distinct construct that could potentially be

explored quantitatively. Importantly, the development of a multi-indicator latent construct of maternal positivity suggests that theoretically, researchers can measure positivity between groups and investigate its potential function. Chapter 2 also found that, like single indicators of positivity, maternal positivity as a latent construct had a negative association with negative outcomes (maternal psychological distress, parenting stress and child behaviour problems). Resilience theory proposes that the reason that some people are able to successfully adapt to a maladaptive situation is because they are more resilient than those that do not adapt well (Masten, 2018; Rutter, 1985). Chapter 2 demonstrated that maternal positivity could potentially be conceptualised as a resilience variable that supported maternal well-being however further investigation was needed before firm conclusions could be made.

Chapter 3 addressed the question of whether maternal positivity is a moderating factor and/ or acts more directly on maternal mental health. Results showed that mothers caring for a child with ID exhibiting behaviour problems and psychopathology demonstrate 'resilience' under conditions of high maternal positivity cross-sectionally. In addition, maternal positivity buffered the impact of child behaviour problems on parenting stress however it had no function longitudinally. From a resilience theory perspective, which aims to understand why some are able to overcome adversity whilst others do not, maternal positivity enables mothers to directly improve their mental health in the face of challenges associated with raising a child with ID but in the short-term only. Thus, maternal positivity should be conceptualised as a resilience variable that has a predominantly compensatory function.

Maternal positivity as a resilience variable indicates that within family disability research, it should be viewed as a multi-domain construct that functions to improve maternal well-being. Chapters 2 and 3 have important theoretical implications and highlights the need for theories centred around coping and resilience to adopt an approach that considers multiple positive psychological states simultaneously. Folkman's theory of stress and coping proposes

that positive affect may enhance psychological and physical resources during stress and protect a person's mental health against the effects of stress. However, these theories have been drawn upon and extended within the context of family disability research. Chapter 2 demonstrated that in addition to general positive feelings (positive affect), positive perceptions specific to parenting and disability strongly reduces maternal stress (psychological distress: $\beta = -.94$, $p = <.001$; parenting stress: $\beta = -.99$, $p= <.001$).

Maternal positivity did not have either a compensatory or protective function between time points therefore could not be conceptualised as a resilience variable over time. Whilst much of the existing ID family research often posit child behaviour problems as the consistent 'risk variable', researchers need to diversify their attention to other potential risk factors. Arguably, maternal positivity could not have moderated the impact of child behaviour problems because they were not found to be significant longitudinal risk variables. Therefore, maternal positivity could not demonstrate a protective function in the longitudinal analysis because longitudinally, child behaviour problems did not place any significant risk to maternal psychological distress or parenting stress. Although only a covariate, maternal employment was the only variable that had a significant (negative) effect on maternal psychological distress longitudinally which implies that maternal unemployment should be considered as a risk variable when assessing how effective maternal positivity is at protecting maternal stress against risk. Alternatively, differential results for cross-sectional and longitudinal data were because the protective role of positivity is only measurable in close association to the timing of the mental health outcome and so the impact of maternal positivity cannot extend into the future.

Previous research on the Down's syndrome (DS) advantage largely focused on negative outcomes for parents and few have attempted to explore whether this is applicable to positive outcomes. Chapter 4 explored the theoretical hypothesis that the DS advantage exists

for positive maternal outcomes in addition to negative maternal outcomes. Initial analysis revealed that the DS advantage was present, indicating that mothers raising children with DS perceive their child to have a more positive impact on themselves and their family, will have a closer relationship with their child and will report lower levels of psychological distress than mothers raising children with an unknown diagnosis of ID. However, upon further analysis, once control variables were accounted for the DS advantage remained only for positive gains. These findings suggest that the reason mothers raising children with DS have better outcomes, with exception to positive gains, is due to external factors and not their child's diagnosis, therefore presenting conflicting results. Whilst the DS advantage remained for one positive outcome (positive gains) it was no longer statistically present for the other (parent-child closeness). Although not evident by means of significance analyses, small effect sizes were still present for all maternal outcomes after controlling for child and maternal variables, offering some support to the DS advantage theory.

Chapter 4 challenges the narrative that having a child with ID is a fundamentally negative experience. Findings further highlight the need to broaden the approach to the DS advantage by addressing the socioeconomic and child-related factors which were shown to interact with maternal psychological distress and child-parent closeness. This would ensure that mothers caring for children with an unknown diagnosis do not have worse outcomes than mothers of children with DS, particularly when it pertains to child-related benefits (positive gains and parent-child closeness).

The approach to group comparison studies involves implementing measurement instruments for all groups and comparing measurement scores to identify potential differences. However, this approach implicitly assumes that an instrument is measuring the same concept across all groups. Chapter 5 demonstrated that the Positive Gains Scale (PGS) does not operate equivalently for mothers of typically developing (TD) children, mothers of

children with a chronic physical health problem (CPHP) and mothers of children with ID indicating that any comparisons of PGS mean scores could, therefore, be invalid. Worryingly it calls into question any conclusions made that have been founded on comparison design studies in which the instrument hasn't been tested for measurement invariance.

Maternal positivity remains a relatively new area of research therefore comparison studies are essential for understanding potential group differences. However, chapter 5 suggests that measurement invariance testing is imperative to inform theory and should be an essential preliminary requirement to ensure that conclusions about group differences are valid.

Methodological limitations

Whilst the research in this thesis makes many unique contributions to the literature surrounding maternal positivity there were several methodological limitations to the studies included in this thesis. Firstly, mothers provided all the data in this study which means there was a problem of source variance. Future research will need to incorporate independent or multiple informant approaches for key constructs (e.g., child behavioural problems and prosocial behaviours) to address source variance.

Secondly, although child-ages ranged from 3 to 18 years old, child age was not accounted for in every study and this could have influenced results. Although a wide age range allows for generalisations to be made across a large group this approach does not account for potential age-related differences and their consequent impact on findings. This was explicitly evident in chapters 3 and 4 in which child-age was associated with parenting stress and child-parent closeness respectively. Future research should consider potential child-age differences and therefore account for age during analysis.

Importantly the studies in this thesis did not permit a developmental perspective and age was only explored as a correlate. The longitudinal data (Chapter 3) were only two time points at a short time apart, thus, developmental effects associated with child age were not examined. This further emphasises the need to explore the function of maternal positivity over a longer time period. Researchers will need to include multiple time points over a longer period of time to account for developmental changes and how they may interact with the way maternal positivity functions for mothers caring for children with ID. It remains unknown whether the latent construct of maternal positivity has a resilience function beyond one year and so this will need to be explored over a longer time frame. Consideration should be made to the possibility that a longer distance between time points could emit different results.

Diagnostic status was not confirmed by a professional or clinical report for all child participants. Therefore, it cannot be professionally confirmed that each child in this thesis had the diagnosis specified. This is particularly important for comparison studies such as those of chapters 4 and 5. Different diagnoses have different characteristics which could impact findings. For example, unknowingly including an undiagnosed child with autism, which is - strongly associated with significantly heightened behaviour problems, within a comparison group of a different ID diagnoses may skew results. Therefore, it is important that researchers make every effort to obtain accurate participant diagnosis to ensure correct conclusions are made.

Although structural equation modelling can demonstrate reciprocal and causal relationships between latent and observed variables there are some limitations to this approach. Fit indices for latent constructs are dependent on the population data therefore it is important to acknowledge that the proposed construct of maternal positivity may not be valid for a different set of mothers. A different set of indicators could be more applicable and more aligned to mothers raising children without disabilities. Therefore, it is important to

acknowledge that positivity may come in varying forms and should not be restricted to a specific set of indicators. It is also important to note that because secondary data were used to create the latent variable, I was limited to a pre-existing set of completed measures to create maternal positivity. The problem with selecting measures from a pool of pre-existing measures is that I was unable to preselect measures I thought would potentially be more applicable to maternal positivity. There are likely other single indicators of positivity that, for example, have strong negative associations with maternal psychopathology that should be explored. Self-efficacy emerged as loading the strongest to the latent variable which suggests that measures focused on parenting and feeling highly efficacious as a parent are best placed to define what maternal positivity looks like for mothers of children with ID.

There are further vulnerabilities when creating a latent construct using multiple measures. For example, participants may respond to different measures variably dependent on the nature of that measure. Arguably, responses by mothers of children with disabilities may be vulnerable to social desirability bias which mainly occurs for items that deal with personally or socially sensitive content. Mothers of children with ID could feel pressure to be viewed by others as highly competent in their parenting due to social pressures and expectations. Parental self-efficacy, for example, could be vulnerable to scores that are too high relative to a person's true score if the respondent wants to be viewed to be more efficacious than they actually are. In addition, a subjective construct such as family satisfaction is subject to change frequently if family relationships are volatile. Therefore, respondents could display a variable pattern of very high scores or very low scores across a latent construct dependent on the nature of the indicators.

In addition to social desirability bias, chapter 5 raised the implications of response styles. A dominant focal topic within disability literature has been on negative rather than positive outcomes. Mothers of disabled children may feel more compelled to emphasise the

positive aspects of raising their child, more so than mothers of non-disabled children, due to the wider negative perception of disability.

Although survey design studies have strong advantages, one being that they can reach a large number of participants in a relatively short time, this methodological approach does have its disadvantages. Self-selection bias occurs when respondents select themselves for a study which is what occurred for the studies included in this thesis. Arguably, those that volunteered to participate in the current studies may have different demographic characteristics and measure responses to those that both dropped out and decided not to participate. For example, the 1000 Families Study was largely advertised online through disability charity websites, online support groups and chat forums. This approach inadvertently excludes potential participants that don't have access to the internet. Furthermore, advertising via charities both online and in person also inadvertently excludes those that may be isolated and not connected to external agencies. Mothers of children with Spina Bifida and/ or Hydrocephalus (chapter 5) were recruited through the register of the Association for Spina Bifida and Hydrocephalus (ASBAH). This register only included families that had contacted the ASBAH for information and/or support. Therefore, given the association between social support and well-being it should be considered that a non-self-selecting sample may have reported differently to measures included in this thesis. A more isolated sample with no access to support may have reported lower scores for well-being measures and lower scores of positivity measures. Avoiding self-selection bias is somewhat challenging for large survey-design studies however including additional methods of recruitment may be one way to reduce it. However, in the case of group comparison design studies, using multiple methods of recruitment may result in vastly different participant numbers if different methods are applied to each group as was the case in Chapter 5. Therefore, to avoid this I would strongly suggest that where possible, researchers should use

multiple methods of recruitment to source participants and to also apply that same approach when recruiting comparison groups.

Finally, an important point to address is the ambiguity of the SES composite variable introduced in chapter 4. This variable was created using four single indicators (household income, hardship, financially managing and education qualifications). It was proposed that low scores indicated low socioeconomic status, however individually they may be indicative of other factors. It is presumed that a low household income is indicative of families having trouble accessing resources and/or enjoying leisure time. However, it should also be considered that low household income is not always indicative of income poverty. Some families with a low income may in fact have a lower cost of living and therefore do have disposable income which they can use to access the resources they need and enjoy leisure activities. Conversely, families that have a high household income may also have a high cost of living which therefore limits their access to resources and support outside the home, thus experiencing income poverty. Similarly, educational qualifications are used as an indicator of SES yet again this may be a misleading approach. More often than not, if a parent needs to give up work to care for a disabled child it will usually be the mother. Therefore, irrespective of the mother's high qualifications and earning potential, if she is not working her qualifications offer minimal insight into the family's socioeconomic status. These are considerations to be made for further research and for the development of future composite variables.

Future research implications

There were many strengths to the empirical studies included in this thesis that have implications for further research. A consensus of what maternal positivity 'looks like' is much needed within family disability literature and this thesis (Chapter 2) has gone some way to do this by presenting a multi-indicator construct that taps onto different domains of

maternal life. Whilst it cannot be concluded that this construct is applicable to all mothers, this thesis does demonstrate that positivity specific to mothers raising children with ID can be conceptualised using multiple indicators and thus should be considered as such in further research and for the development of quantitative instruments that measure positivity. As such, there is a further need to broaden the theoretical approach to stress and coping models by defining positivity as a multi-domain construct.

Future research into latent constructs of positivity should consider including additional single indicators or replacing some of the weaker ones. Parental self-efficacy was the strongest indicator to load to the construct indicating that a mother's confidence in their ability to parent their child with ID is particularly important when considering maternal positivity. Parenting self-efficacy pertains to one feeling they have control over their child's behaviour and their ability to manage it (Hastings & Brown, 2002). Future research should explore other indicators that pertain to mothers being the agent of control opposed to being passive. In addition, support has been found to be associated with positive outcomes for mothers raising children with ID (Hassall, Rose & McDonald, 2005; Meppelder, Hodes, Kef & Schuengel, 2015; Skok, Harvey & Reddihough, 2006) therefore perceived satisfaction with social support could also be explored as a potential maternal positivity indicator.

Chapter 2 demonstrated that potentially maternal positivity could be developed as a quantifiable measure. Further research will need to be conducted to create a valid instrument that can reliably measure maternal positivity. Guided by the latent construct, items that measure general positivity, positivity specific to parenting and raising a child with ID should be included in the development of a maternal positivity measure.

Developing a quantifiable measure of maternal positivity is important to clearly understand how it functions and whether there are differences between diagnostic groups.

Potential differences between groups would help to inform interventions. Importantly, if a reliable measure of maternal positivity is developed, additional investigation would be required to determine whether this measure is equivalent across all comparison groups. If mean scores cannot be compared chapter 5 demonstrated that it could still be possible to compare single items, and this may offer valuable insight into potential group differences in further research.

Chapter 3 showed that maternal positivity has both a protective and compensatory function cross-sectionally but neither function longitudinally. It was previously discussed that the lack of longitudinal findings could have been due to study design which is why future studies should look to extend the time between initial data collection and subsequent data collection to determine whether maternal positivity can ameliorate the impact child behaviour problems have on maternal mental health. Furthermore, alternative risk factors should be considered given that maternal unemployment proved to be a longitudinal risk factor and child factors were not. Researchers should widen their approach to potential risk factors and broaden their focus to include non-child related variables.

Chapter 2 identified negative associations between maternal positivity and poor maternal mental health therefore suggesting that as one construct increases the other decreases. Yet it remains unknown whether poor maternal mental health has a direct effect on maternal positivity and thus the five key areas that collectively make up the latent construct of maternal positivity. It is well established within the literature that mothers raising a child with ID have poorer mental health than mothers raising typically developing children. Therefore, further research is needed to identify whether the poor mental health experienced by mothers of children with ID has a direct effect on these key areas identified collectively as maternal positivity. Importantly, child prosocial and challenging behaviours also had significant associations with maternal positivity, yet these relationships were not explored

any further within the thesis. Therefore, further research is recommended to explore the potential impact of maternal positivity on child behaviours.

This thesis evidenced that the Down's syndrome advantage was largely attributed to external factors such as child adaptive skills and child age. These findings indicate that further research is needed to explore whether there are additional variables, not included in the study that may also have an effect on maternal outcomes. It is equally important to know why external factors have differential effects on varying maternal outcomes. For example, socio-economic status interacted with maternal psychological distress but not child-parent closeness. In this instance the DS advantage still remained for positive gains however it could indicate that significant covariates were overlooked. It has been shown that positive gains has significant associations with parental empowerment (Minnes & Weiss, 2014) mindfulness and acceptance (Jones, Hastings, Totsika, Keane & Rhule, 2014) therefore further research will need to account for maternal psychological constructs and their potential interaction with perceived positive gains of their child with ID and consider them as potential covariates in further research.

The psychological distress seemingly associated with ID diagnoses was actually accounted for by a socioeconomic disadvantage. Researchers have found that the opportunity for parents of children with ID, particularly mothers, to work and thus generate an income is limited by inflexible employment conditions and access to childcare (McConnell & Savage, 2015). Findings may indicate unequal access to informal and/ or formal childcare resources thus perhaps explaining in part why there were differences between groups. Thus, it is worth investigating whether maternal positivity could moderate the adverse impact of low income on maternal mental health.

Chapter 4 also touched on the prospect of extending the findings by dividing the heterogeneous 'ID' group into multiple homogeneous comparison groups separated by a specific ID diagnosis. This could potentially identify whether the covariates included in the study interact with the PGS differentially across ID diagnoses. Consequently, such findings could identify vulnerable groups and accurately inform clinicians about who to target for specific interventions. For example, if it were found that child age as a covariate had a negative interaction with positive gains for mothers of children with Cerebral Palsy, clinicians would know that mothers of older children with cerebral palsy were vulnerable to perceiving their child less positively than mothers of younger children. Such information would enable clinicians to implement interventions focused on tackling the factors that cause mothers of older children with cerebral palsy to perceive their child less positively.

Practical implications

When child behaviour problems are conceptualised as stressors there is the tendency for interventions aimed at improving maternal well-being to focus on the child (i.e., reducing behaviour problems). This thesis suggests that high scores of the five indicators that constitute 'maternal positivity' are vital for the reduction of maternal psychological stress and for protecting mothers raising children with ID from the adverse effects child behaviour problems have on parenting stress. Practitioners would need to develop a programme which works to purposefully increase all dimensions of maternal positivity. This somewhat presents an alternative intervention if those that are child-focused are unsuccessful (i.e., child behaviour therapies). In addition, increasing maternal positivity could potentially reduce child behaviour problems and child psychopathology however this would need to be explicitly tested for.

This thesis has implications for the existing narrative surrounding ID and diagnostic differences. The wider view has been that raising a child with ID is a negative experience

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with poor outcomes for the parents and the child whilst parents of children with DS have comparatively better outcomes. Chapter 4 indicates that once methods for improving external factors have been implemented mothers will have equally positive experiences. Therefore, such methods need to be explored and developed to ensure that parents of children with an unknown ID diagnosis do not have worse outcomes than mothers raising a child with DS.

However, chapter 4 demonstrated that mothers of children with DS do appear to view their child more positively than mothers of children with an unknown ID diagnosis. Arguably this information should be more widely available, particularly with the increased availability of non-invasive prenatal testing (NIPT). Knowing that raising a child with Down's syndrome is often a positive experience for themselves and their family may better inform women pregnant with a baby with Down's syndrome.

There needs to be a common consensus within the literature of what maternal positivity looks like so that an appropriate measure can be developed and implemented in future studies to widen our understanding of maternal positivity. This thesis proposes that maternal positivity should be conceptualised as a multi-domain construct. Although this construct of maternal positivity is only applicable to the studied population its development does indicate that multi-indicator constructs should be considered when attempting to measure maternal positivity or understand its function thus the development of a quantifiable measure is proposed. However, Chapter 5 demonstrated the importance of measurement invariance and so clinicians will need to determine prior to use whether an instrument that measures maternal positivity is appropriate for their client group otherwise they may not be measuring the construct they had intended to. This does not only apply to instruments that measure positive constructs. Clinicians must be mindful that screening tests such as the HADS may not capture anxiety and depression for all patient groups in the same way, particularly if comparisons are going to be made.

Practitioners could also encourage patients that are experiencing difficult and upsetting situations to focus on the positive aspects of what is happening. According to Folkman and Moskowitz, (2000) this approach would evoke positive emotions which in turn would allow a person to experience positive well-being.. It would be of further interest to explore whether mothers of children with DS also report lower levels of stress and whether this could also be explained by the same covariates included in chapter 4. If there are no group differences in reported levels of stress yet differences are still evident for positive gains, Folkman's theory of stress and coping may be best placed to explain this, in that mothers of children with DS are more inclined to focus on the positive aspects of their situation thus having more positive outcomes. Perhaps it is because mothers of children with DS have access to information about their child's condition and prognosis that allows them to focus on the positive aspects of their experience. In contrast, mothers of children with an unknown diagnosis have very limited information about their child's prognosis. More research needs to be done in order to understand more about the various syndromes that are yet to be named in order for more information to be readily available for parents.

Conclusion

Finally, this thesis has demonstrated that the general negative narrative surrounding raising a child with ID is outdated. Mothers raising children with ID are positive about their child and variations in reported levels of positivity appear to be largely grounded in factors outside of their child's diagnosis. Importantly, this thesis proposes that increasing positivity across multiple domains of maternal life will reduce maternal stress which have implications for practice and theory.

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Appendices

Appendix I

Cerebra 1,000 Families study
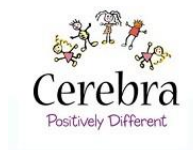
Primary caregiver survey

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 ONE	✓
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 ONE	✓
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 ONE	✓
Male	
Female	

Questions about you

14. Please indicate your relationship to the child with intellectual disability.

Please select ONE	✓
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 ONE	✓
Male	
Female	
Trans	
Prefer not to answer	

16. How would you describe your ethnicity?

Please select ONE	✓
Asian/Asian British: Indian	
Asian/Asian British: Pakistani	
Asian/Asian British: Bangladeshi	
Asian/Asian British: Chinese	
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)	

17. Please select the highest level of your educational qualifications

Please select ONE	✓
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 ONE	✓
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 ONE	✓
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 ONE	✓
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 <u>weekly</u> 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 ALL 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)	

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 ONE	✓
Yes	
No	

30. Does your child with intellectual disability currently have epileptic seizures?

Please select ONE	✓
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 got to Question 35)	

35. Please state the approximate number of hours that your child lives with you on a week

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 ONE 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 ANY 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 ONE 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>	✓
Married and living with spouse/civil partner (Please answer questions 42 and 43)	
Living with partner (Please answer questions 42 and 43)	
Divorced/Separated/Single/Widowed/Not currently living with partner (Please go to question 44)	

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>	✓
Never	
Less than once a week	
Once a week	
Several times a week	
Once a day	
More than once a day	
Can't say	

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			

I am satisfied with the way my family talks over things with me and shares problems with me			
I am satisfied that my family accepts and supports my wishes to take on new activities or directions			
I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love			
I am satisfied with the way my family and I share time together			

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
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Relationship with your child with intellectual disability

We would now like to ask you about your relationship with your <u>child with intellectual disability</u>.

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 <u>one</u> appropriate answer for each item.

Please select <u>ONE answer per</u> <u>statement</u>	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 ONE 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	185				
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 ONE 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 <u>closest in age</u> to your child with intellectual disability.

50. Age of sibling in years and months

Years ☐   **Months** ☐

51. Sibling gender

Please select <u>ONE</u>	✓
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 <u>ONE</u>	✓
No	
Yes (Please also answer question 54)	

53. Does this sibling live in the same household as your child with intellectual disability?

Please select <u>ONE</u>	✓
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
██████████████████			
█████████████████████			
█████████████████████████ ████████			

████████████████████████ ████████			
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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 answer per</u> 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

Appendix II

The Satisfaction with Life Short Scale

Below are five statements that you may agree or disagree with. Using the 1-7 scale below indicate your agree with each item by placing the appropriate number on the line preceding that item. Please be open and honest in your responding.

- 7- Strongly agree
- 6- Agree
- 5- Slightly agree
- 4- Neither agree nor disagree
- 3- Slightly disagree
- 2- Disagree
- Strongly disagree

1. _____ In most ways my life is close to ideal.
2. _____ The conditions of my life are excellent.
3. _____ I am satisfied with my life.
4. _____ So far I have gotten the important things I want in life.
5. _____ If I could live my life over, I would change almost nothing.

Appendix III

Family Satisfaction Scale

The following questions are about how satisfied **you are** with these aspects of your **family relationship**. Please tick the box that you feel is most appropriate for each statement.

		Very Dissatisfied	Somewhat Dissatisfied	Generally Satisfied	Very Satisfied	Extremely Satisfied
1.	The degree of closeness between family members	☐	☐	☐	☐	☐
2.	Your family's ability to cope with stress	☐	☐	☐	☐	☐
3.	Your family's ability to be flexible	☐	☐	☐	☐	☐
4.	Your family's ability to share positive experiences.	☐	☐	☐	☐	☐
5.	The quality of communication between family members	☐	☐	☐	☐	☐
6.	Your family's ability to resolve conflicts.	☐	☐	☐	☐	☐
7.	The amount of time you spend together as a family.	☐	☐	☐	☐	☐
8.	The way problems are discussed.	☐	☐	☐	☐	☐
9.	The fairness of criticism in your family.	☐	☐	☐	☐	☐
10.	Family members concern for each other.	☐	☐	☐	☐	☐

Appendix IV

The Positive and Negative Affect Schedule (PANAS)

This scale consists of a number of words that describe different feelings and emotions. Read each item and then click on the response on the dropdown list next to the word and select one of the responses. **Indicate to what extent you feel this way right now, that is, at the present moment.**

	Very slight of not at all	A little	Moderate	Quite a bit	Extremely
1. **Interested**	1	2	3	4	5
2. **Excited**	1	2	3	4	5
3. **Strong**	1	2	3	4	5
4. **Enthusiastic**	1	2	3	4	5
5. **Proud**	1	2	3	4	5
6. **Alert**	1	2	3	4	5
7. **Inspired**	1	2	3	4	5
8. **Determined**	1	2	3	4	5
9. **Attentive**	1	2	3	4	5
10. **Active**	1	2	3	4	5

Appendix V

Parenting Efficacy

Please circle either number 1 or number 7. If your views are somewhere in between the two end points, please select a position on the scale that reflects where you feel your views should be placed. Please select a response for each of the questions.

How confident are you in parenting your child with special needs?

1   2   3   4   5   6   7

Not at all confident   Very confident

How difficult do you personally find it to parent your child with special needs?

1   2   3   4   5   6   7

Very difficult   Not at all difficult

To what extent do you feel that the way you parent your child with special needs has a positive effect?

1   2   3   4   5   6   7

Has no positive effect at all   Has a very positive effect

How satisfied are you with the way you parent your child with special needs?

1   2   3   4   5   6   7

Not satisfied at all   Very satisfied

To what extent do you feel in control when parenting your child with special needs?

1   2   3   4   5   6   7

Not in control   Very much in control

Appendix VI

[1]Questionnaire on Resources and Stress (QRS)-Full

This questionnaire deals with your feelings about a child in your family. There are many blanks on the questionnaire. Imagine the child's name filled in on each blank. Give your honest feelings and opinions. Please answer all of the questions, even if they do not seem to apply. If it is difficult to decide True (T) or False (F), answer in terms of what you or your family feel or do *most* of the time. Sometimes the questions refer to problems your family does not have. Nevertheless, they can be answered True or False, even then. Please begin. Remember to answer all of the questions.

1. _____ doesn't communicate with others of his/her age group.   T   F
2. Other members of the family have to do without things because of _____.   T   F
3. Our family agrees on important matters.   T   F
4. I worry about what will happen to _____ when I can no longer take care of him/her.   T   F
5. The constant demands for care for _____ limit growth and development of someone else in our family.   T   F
6. _____ is limited in the kind of work he/she can do to make a living.   T   F
7. I have accepted the fact that _____ might have to live out his/her life in some special setting (e.g. an institution or group home).   T   F
8. _____ can feed himself/herself.   T   F
9. I have given up things I have really wanted to do in order to care for _____.   T   F
10. _____ is able to fit into the family social group.   T   F
11. Sometimes I avoid taking _____ out in public.   T   F
12. In the future, our family's social life will suffer because of increased responsibilities and financial stress.   T   F
13. It bothers me that _____ will always be this way.   T   F
14. I feel tense whenever I take _____ out in public.   T   F
15. I can go visit friends whenever I want.   T   F
16. Taking _____ on vacation spoils pleasure for the whole family.   T   F
17. _____ knows his/her own address.   T   F
18. The family does as many things together now as we ever did.   T   F
19. _____ is aware who he/she is.   T   F
20. I get upset with the way my life is going.   T   F
21. Sometimes I feel very embarrassed because of _____.   T   F
22. _____ doesn't do as much as he/she should be able to do.   T   F
23. It is difficult to communicate with _____ because he/she has difficulty understanding what is being said to him/her.   T   F
24. There are many places where we can enjoy ourselves as a family when _____ comes along.   T   F
25. _____ is over-protected.   T   F
26. _____ is able to take part in games or sports.   T   F

27. _____ has too much time on his/her hands.   T F

28. I am disappointed that _____ does not lead a normal life.   T F

29. Time drags for _____, especially free time   T F

30. _____ can't pay attention very long.   T F

31. It is easy for me to relax.   T F

32. I worry about what will be done with _____ when he/she gets older.   T F

33. I get almost too tired to enjoy myself.   T F

34. One of the things I appreciate about _____ is his/her confidence.   T F

35. There is a lot of anger and resentment in our family.   T F

36. _____ is able to go to the bathroom alone.   T F

37. _____ cannot remember what he/she says from one moment to the next.   T F

38. _____ can ride a bus.   T F

39. It is easy to communicate with _____.   T F

40. The constant demands to care for _____ limit my growth and development.   T F

41. _____ accepts himself/herself as a person.   T F

42. I feel sad when I think of _____.   T F

43. I often worry about what will happen to _____ when I no longer can take care of him/her.   T F

44. People can't understand what _____ tries to say.   T F

45. Caring for _____ puts a strain on me.   T F

46. Members of our family get to do the same kinds of things other families do.   T F

47. _____ will always be a problem to us.   T F

48. _____ is able to express his/her feelings to others.   T F

49. _____ has to use a bedpan or a diaper.   T F

50. I rarely feel blue.   T F

51. I am worried much of the time.   T F

52. _____ can walk without help.   T F

[1]. Items used to measure 'Parenting Stress': 2,3,5,9,10,12,15,16,18,24,35,40,42,45,46

Appendix VII

THE NISONGER CHILD BEHAVIOR RATING FORM

TIQ VERSION (NCBRF–*TIQ*)

PARENT VERSION

Child's Name: _____   Child's Date of Birth: _____

/_____ /_____

                                            month   day

year

Rater's Name: _____   Date of Rating: _____

/_____ /_____

                                            month   day

year

Relation of Rater to Child: parent [1]   other [9]:

_____

                                          (please specify)

Please describe any special circumstances or mediating factors that may have affected the child's behavior inthe recent past (the last month or two) or prevented you from making complete ratings.

POSITIVE SOCIAL. Please describe the child's behavior as it was at home over the last month.

198

MATERNAL POSITIVITY IN MOTHERS RAISING CHILDREN WITH ID

IN THE LAST MONTH, THIS CHILD HAS:	Not True [0]	Somewhat or Sometimes True [1]	Very or Often True [2]	Completely or Always True [3]
1. Accepted redirection	☐	☐	☐	☐
2. Expressed ideas clearly	☐	☐	☐	☐
3. Followed rules	☐	☐	☐	☐
4. Initiated positive interactions	☐	☐	☐	☐
5. Participated in group activities	☐	☐	☐	☐
6. Resisted provocation, was tolerant	☐	☐	☐	☐
7. Shared with or helped others	☐	☐	☐	☐
8. Stayed on task	☐	☐	☐	☐
9. Was cheerful or happy	☐	☐	☐	☐
10. Was patient, able to delay	☐	☐	☐	☐

PAGE 1   **(OVER)**

Nisonger CBRF–*TIQ*: Parent

Appendix VIII

CHILD-PARENT RELATIONSHIP SCALE
Robert C. Pianta

Child:_____   Age:_____

Parent:_____

Please reflect on the degree to which each of the following statements currently applies to your relationship with your child. Using the scale below, circle the appropriate number for each item.

Definitely does not apply 1	Not really 2	Neutral, not sure 3	Applies somewhat 4	Definitely applies 5

1. I share an affectionate, warm relationship with my child.	1	2	3	4	5
2. My child and I always seem to be struggling with each other.	1	2	3	4	5
3. If upset, my child will seek comfort from me.	1	2	3	4	5
4. My child is uncomfortable with physical affection or touch from me.	1	2	3	4	5
5. My child values his/her relationship with me.	1	2	3	4	5
6. My child appears hurt or embarrassed when I correct him/her.	1	2	3	4	5
7. My child does not want to accept help when he/she needs it.	1	2	3	4	5
8. When I praise my child, he/she beams with pride.	1	2	3	4	5
9. My child reacts strongly to separation from me.	1	2	3	4	5
10. My child spontaneously shares information about himself/herself.	1	2	3	4	5
11. My child is overly dependent on me.	1	2	3	4	5
12. My child easily becomes angry at me.	1	2	3	4	5
13. My child tries to please me.	1	2	3	4	5
14. My child feels that I treat him/her unfairly.	1	2	3	4	5
15. My child asks for my help when he/she really does not need help.	1	2	3	4	5
16. It is easy to be in tune with what my child is feeling.	1	2	3	4	5
17. My child sees me as a source of punishment and criticism.	1	2	3	4	5
18. My child expresses hurt or jealousy when I spend time with other children.	1	2	3	4	5
19. My child remains angry or is resistant after being disciplined.	1	2	3	4	5
20. When my child is misbehaving, he/she responds to my look or tone of voice.	1	2	3	4	5

21. Dealing with my child drains my energy.	1	2	3	4	5
22. I've noticed my child copying my behavior or ways of doing things.	1	2	3	4	5
23. When my child is in a bad mood, I know we're in for a long and difficult day.	1	2	3	4	5
24. My child's feelings toward me can be unpredictable or can change suddenly.	1	2	3	4	5
25. Despite my best efforts, I'm uncomfortable with how my child and I get along.	1	2	3	4	5
26. I often think about my child when at work.	1	2	3	4	5
27. My child whines or cries when he/she wants something from me.	1	2	3	4	5
28. My child is sneaky or manipulative with me.	1	2	3	4	5
29. My child openly shares his/her feelings and experiences with me.	1	2	3	4	5
30. My interactions with my child make me feel effective and confident as a parent.	1	2	3	4	5

CHILD-PARENT RELATIONSHIP SCALE

Scoring Guide

Sum the items as noted; each question has a score from 1 – 5.

To establish the mean, divide the sum by the number of questions in that section.

Conflicts

2   seem to be struggling with each other

12   easily becomes angry with me

14   feels I treat him/her unfairly

17   sees me as a source of punishment
18   hurt when I spend time with other child
19   remains angry after discipline
21   dealing with child drains energy

23   bad day when child wakes up in a bad mood
24   feelings toward me can be unpredictable
25   uncomfortable with how child and I get along
27   whines when he/she wants something
28   sneaky or manipulates me

Positive aspects of relationship (closeness)

1   an affectionate relationship

3   will seek comfort from me if upset

5   values his/her relationship with me

8   beams with pride when praised

10   spontaneously shares information

13   tries to please me

16   easy to be in tune with child=s feelings

22   copies my behavior

29   openly shares feelings with me

30   interactions make me feel effective

Dependence

6   appears hurt when corrected

9   reacts strongly to separation from me

11   overly dependent on me

26   think about child when not together

Appendix IX

Positive Gains Scale

Please select <u>ONE answer per statement</u>	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					

Appendix X

Kessler K6

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 answer per statement</u>	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?					

Appendix XI

SES composite Variable

Please select the highest level of your educational qualifications

Please select ONE	✓
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	

What is your total <u>weekly</u> 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 ONE	✓
£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	

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	

Appendix XII
DEVELOPMENTAL BEHAVIOUR CHECKLIST

(DBC-P)

Some children with developmental delay have problems with their emotions and behaviour. These can sometimes be a problem for their carers.

By completing this checklist, you will help us learn more about these problems. This will assist us to know how the person might respond to help.

Name of Child or Teenager:

Date of Birth/Age:.

Sex:

Person Completing Form:

Relationship to Child:

Date Completed:

Is the Child: (please circle)   Unable to see / unable to hear   Unable to speak/ speaks very little

Unable to use arms / legs   Subject to other serious medical condition

Please describe:

What does he/she do best?

_____

What do other people like about him/her?

_____

What are his/her favourite activities?

_____

Is there anything you feel he/she does as well or better than others?

_____

Have you sought help for any behaviour or emotional problems, apart from slow development, of the child or teenager in your care?   Yes / No

If so, from whom?

_____

Please continue over the page ✑

_____

Office Use Only

Code Number: _____

Developmental Level (circle one only)

Items   Stewart L. Einfeld, Bruce J. Tonge, 1989

Instructions   1981 T.M. Achenbach. modified, with permission   **Revised subscales 2002**

-2-

Many of the following behaviours may not apply to the child or teenager in your care. For each item that does describe the person in your care, now or within the <u>past six months</u>, please circle the **2** if the item is **very true** or **often true**.   Circle **1** if the item is **somewhat or sometimes true** of your child. If the item is **not true** of your child circle the **0**.

0 = not true as far as you know   1 = somewhat or sometimes true   2 = very true or often true

If your child is unable to perform an item, circle the **0.** For example, if your child has no speech, then for the item "Talks too much or too fast" circle the **0**

Underline any you are particularly concerned about

Please Circle

0   1   2   Appears depressed, downcast or unhappy

0   1   2   Avoids eye contact. Won't look you straight in the eye.

0   1   2   Aloof, in his/her own world.

0   1   2   Abusive. Swears at others.

0   1   2   Arranges objects or routine in a strict order.

_____

0   1   2   Bangs head.

0   1   2   Becomes over-excited.

0   1   2   Bites others.

0   1   2 Cannot attend to one activity for any length of time, poor attention span.

0   1   2   Chews or mouths objects, or body parts.

0   1   2   Cries easily for no reason, or over small upsets.

0   1   2   Covers ears or is distressed when hears particular sounds.

Please describe: _____

0   1   2   Confuses the use of pronouns e.g. uses "you" instead of "I".

0   1   2   Deliberately runs away.

0   1   2   Delusions: has a firmly held belief or idea that can't possibly be true.

Please describe: _____

0 1 2   Distressed about being alone.

0 1 2   Doesn't show affection.

0 1 2   Doesn't respond to others' feelings, e.g. shows no response if a family member is crying.

0 1 2   Easily distracted from his/her task, e.g. by noises.

0 1 2   Easily led by others.

0 1 2   Eats non-food items e.g. dirt, grass, soap.

0 1 2   Excessively distressed if separated from familiar person.

0 1 2   Fears particular things or situations, e.g. the dark or insects.

Please describe: _____

0 1 2   Facial twitches or grimaces.

0 1 2   Flicks, taps, twirls objects repeatedly.

0 1 2   Fussy eater or has food fads.

0 1 2   Gorges food. Will do anything to get food e.g. takes food out of garbage bins or steals food.

0 1 2   Gets obsessed with an idea or activity.

Please describe: _____

0 1 2   Grinds teeth.

0 1 2   Has nightmares, night terrors or walks in sleep.

Please be sure you have answered all items

Continue next page

Office Use Only   Subscales

TBPS

-3-
0 = not true as far as you know   1 = somewhat or sometimes true   2 = very true or often true
Underline any you are particularly concerned about

Please Circle

    0   1   2   Has temper tantrums, e.g. stamps feet, slams doors

0   1   2   Hides things.

  0 1   2 Hits self or bites self.

  0   1   2 Hums, whines, grunts, squeals or makes other non-speech noises.

  0   1   2   Impatient.

  0   1   2 Inappropriate sexual activity with another.

  0   1   2   Impulsive, acts before thinking.

  0   1   2   Irritable.

  0   1   2   Jealous.

  0   1   2   Kicks, hits others.

  0   1   2 Lacks self-confidence, poor self-esteem.

  0   1   2 Laughs or giggles for no obvious reason.

  0   1   2   Lights fires.

  0   1   2   Likes to hold or play with an unusual object, e.g. string, twigs; overly fascinated with something, e.g. water.

      Please describe: _____

  0   1   2 Loss of appetite.

  0   1   2 Masturbates or exposes self in public.

0   1   2 Mood changes rapidly for no apparent reason.

0   1   2 Moves slowly, underactive, does little, e.g. only sits and watches others.

0   1   2   Noisy or boisterous.

0   1   2 Overactive, restless, unable to sit still.

0   1   2   Overaffectionate.

0   1   2 Overbreathes, vomits, has headaches or complains of being sick for no physical reason.

0   1   2   Overly attention-seeking.

0   1   2 Overly interested in looking at, listening to or dismantling mechanical things e.g. lawnmower, vacuum cleaner.

0   1   2   Poor sense of danger.

0   1   2   Prefers the company of adults or younger children. Doesn't mix with his/her own age group.

0   1   2 Prefers to do things on his/her own. Tends to be a loner.

0 1   2   Preoccupied with only one or two particular interests.
Please describe:

_____

0   1   2 Refuses to go to school, activity centre or workplace.

0   1   2 Repeated movements of hands, body, head or face e.g. handflapping or rocking.

0   1   2 Resists being cuddled, touched or held.

0   1   2 Repeats back what others say like an echo.

0   1   2   Repeats the same word or phrase over and over. **0   1   2**   Smells, tastes, or licks objects.

0   1   2   Scratches or picks his/her skin. **0   1   2**   Screams a lot.

Please be sure you have answered all items

Office Use Only

Subscales

TBPS

-4-

0 = not true as far as you know   1 = somewhat or sometimes true   2 = very true or often true
Underline any you are particularly concerned about Please Circle

0   1   2   Sleeps too little. Disrupted sleep.

0   1   2   Stares at lights or spinning objects.

0   1   2   Sleeps too much.

0   1   2   Soils outside toilet though toilet trained. Smears or plays with faeces.

0   1   2   Speaks in whispers, high pitched voice, or other unusual tone or rhythm.

0   1   2   Switches lights on and off, pours water over and over; or similar repetitive activity.

Please describe: _____

0   1   2   Steals.

0   1   2   Stubborn, disobedient or unco-operative.

0   1   2   Shy.

0   1   2   Strips off clothes or throws away clothes.

0   1   2   Says he/she can do things that he/she is not capable of.

0   1   2   Stands too close to others.

0   1   2   Sees, hears, something which isn't there. Hallucinations.

Please describe: _____

0   **1**   **2**   Talks about suicide.

0   **1**   **2**   Talks too much or too fast.

0   **1**   **2** Talks to self or imaginary people or objects

0   **1**   **2**   Tells lies.

0   **1**   **2**   Thoughts are unconnected. Different ideas are jumbled together with
          meaning   difficult to follow.

0   **1**   **2**   Tense, anxious, worried.

0   **1**   **2**   Throws or breaks objects.

0   **1**   **2**   Tries to manipulate or provoke others.

0   **1**   **2**   Underreacts to pain.

0   **1**   **2**   Unrealistically happy or elated.

0   **1**   **2** Unusual body movements, posture, or way of walking.

          Please describe: _____

0   **1**   **2** Upset and distressed over small changes in routine or environment.

          Please describe: _____

0   **1**   **2** Urinates outside toilet, although toilet trained.

0   **1**   **2**   Very bossy.

0   **1**   **2**   Wanders aimlessly.

0   **1** **2**   Whines or complains a lot.

          Please write in any problems your child has that were not listed above
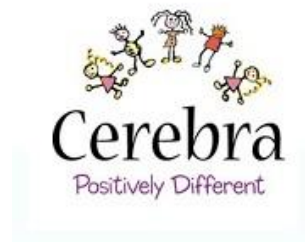
0   **1**   **2**   _____
                       _____
0   **1**   **2**   _____

0   **1**   **2**

0   1   2   Overall, do you feel your child has problems with feelings or behaviour, in addition   to problems with development? If not, please circle the **0**. If so, but they're minor,   please circle the **1.** If they're major problems, please circle the **2**.

Please be sure you have answered all item

Appendix XIII

Primary caregiver study information

1,000 Families study

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 <u>20 minutes</u> 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

Appendix XIV
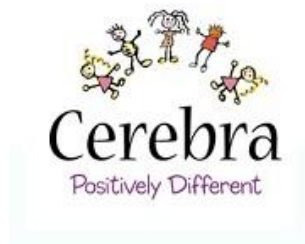
1,000 Families study

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.

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Appendix XV

NHS
Health Research Authority

NRES Committee West Midlands - South Birmingham

Royal Standard Place

Nottingham

NG1 6FS

Tel: 0115 883 9428

11 Septembe**r 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:

Document	Version	Date
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

PP. *[signature]*

Professor Simon Bowman Chair

Email: nrescommittee.westmidlands-southbirmingham@nhs.net

Enclosures: After ethical review – guidance for researchers

Copy to: *Mrs Jane Prewett*

Appendix XVI

7th July 2015

Warwick
Medical School

Prof Richard Hastings
CEDAR
University of Warwick
Coventry
CV4 7AL
United Kingdom

Project Title: The 1000 Families Study
Chief Investigator: Richard Hastings
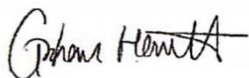**Our Ref:** REGO-2015-1604

Dear Professor Hastings,

I confirm that the University of Warwick will act as Research Sponsor for the above project, in accdordance 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

Dean's Office & Professional Support Services
Warwick Medical School
A010 Medical School Building
The University of Warwick
Coventry
CV4 7AL

T: + 44 (0) 24 7615 1827
E: G.J.Hewitt@warwick.ac.uk
W: www.warwick.ac.uk/wms

Medical School Building

The University of Warwick
Coventry CV4 7AL United Kingdom
Tel: +44 (0)24 7657 4880
Fax: +44 (0)24 7652 8375

THE UNIVERSITY OF
WARWICK

Appendix XVII

NHS
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:

Document	Version	Date
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

Appendix XVIII

ANNUAL PROGRESS REPORT TO MAIN RESEARCH ETHICS COMMITTEE

(For all studies except clinical trials of investigational medicinal products)

To be completed in typescript and submitted to the main REC by the Chief Investigator. For questions with Yes/No options please indicate answer in bold type.

1. Details of Chief Investigator

Name:	Professor Richard Hastings
Address:	CEDAR University of Warwick Coventry CV4 7AL
Telephone:	024 76 522197
E-mail:	R.Hastings@warwick.ac.uk
Fax:	

2. Details of study

Full title of study:	**The 1,000 families study: Well-being in families of children with intellectual disability**
Name of main REC:	West Midlands – South Birmingham
REC reference number:	**15/WM/0267**
Date of favourable ethical opinion:	11 September 2015
Sponsor:	University of Warwick

3. Commencement and termination dates

Has the study started?	*Yes*
If yes, what was the actual start date?	1/11/15
If no, what are the reasons for the study not commencing? What is the expected start date?	
Has the study finished? *If yes, complete and submit "Declaration of end of study" form, available at http://www.nres.npsa.nhs.uk/applications/after-ethical-review/endofstudy/*	*No*

If no, what is the expected completion date? *If you expect the study to overrun the planned completion date this should be notified to the main REC for information.*	**31/12/19**
If you do not expect the study to be completed, give reason(s)	

4. Registration

Is the study a 'clinical trial'? (Defined as first 4 categories on the IRAS filter page) (For CTIMP please use CTIMP progress reporting template)	*No*
Is the study registered on a publically accessible database? (Registration of clinical trials is a condition of approval for studies approved after 30 September 2013)	No

<table>
<tr><td colspan="2">

If yes, please provide the name of the database and the registration number

Database:

Registration number:
</td></tr>
<tr><td colspan="2">

If no:

    a. What is the reason for non-registration?

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.

    b. What are your intentions for registration?
</td></tr>
</table>

5. Site information

Do you plan to increase the total number of sites proposed for the study? If yes, how many sites do you plan to recruit?	No

6. Recruitment of participants

In this section, "participants" includes those who will not be approached but whose samples/data will be studied.

Number of participants recruited:	*Proposed in original application:* Total N proposed up to 4000 individual parents (2000 families maximum) *Actual number recruited to date:* 1035 primary carers (mainly mothers). Final information about numbers of parents recruited to the first phase of this study will be available before the next annual report once internet survey responses have been fully cleaned and checked.
Number of participants completing trial:	*Actual number completed to date:* N/A this is not a trial
Number of withdrawals from study to date due to: (a) withdrawal of consent (b) loss to follow-up (c) death (where not the primary outcome) Total study withdrawals:　　0	
*Number of treatment failures to date (prior to reaching primary outcome) due to: (a) adverse events (b) lack of efficacy Total treatment failures: * Applies to studies involving clinical treatment only	
Have there been any serious difficulties in recruiting participants?	No
If Yes, give details:	

Do you plan to increase the planned recruitment of participants into the study?	No
Any increase in planned recruitment should be notified to the main REC as a substantial amendment for ethical review.	

7. Safety of participants

Have there been any related and unexpected serious adverse events (SAEs) in this study?	No
Have these SAEs been notified to the Committee? *If no, please submit details with this report and give reasons for late notification.*	Not applicable
Have any concerns arisen about the safety of participants in this study? *If yes, give details and say how the concerns have been addressed.*	*No*

8. Amendments

Have any substantial amendments been made to the trial during the year?	Yes
If yes, please give the date and amendment number for each substantial amendment made.	Amendment number SA1 (extending age range of children in the inclusion criteria), approved 7 March 2017

9. Serious breaches of the protocol

Have any serious breaches of the protocol occurred during the year? *If Yes, please enclose a report of any serious breaches not already notified to the REC.*	*No*

10. Other issues

Are there any other developments in the study that you wish to report to the Committee?	*No*
Are there any ethical issues on which further advice is required? *If yes to either, please attach separate statement with details.*	*No*

11. Declaration

Signature of Chief Investigator:	*RP Hastings*
Print name:	Professor Richard Hastings
Date of submission:	5 September 2017

Appendix XIX

Overview of sources of data for each study

Chapter	Primary or Secondary use of Data?	Data source	Role in designing the analytical plan
Chapter 2/ Study 1	Secondary	Secondary data were extracted from the Special Needs and Families Project (SNFP). *Hastings, R. P., Beck, A., & Hill, C. (2005). Positive contributions made by children with an intellectual disability in the family: Mothers' and fathers' perceptions. Journal of Intellectual Disabilities, 9, 155-165*	Structural equation modelling was utilised in this study using AMOS. My second supervisor suggested this method of analysis. After I researched this methodology, I believed it to be the strongest method to address the research question. In addition, AMOS produced figures which I found useful for aiding my interpretation of the results.
Chapter 3/ Study 2	Secondary	Same as above	In part, this study aimed to identify whether Maternal Positivity could moderate the impact of child behaviour on maternal outcomes. Of all the possible methods for testing moderation, I decided to use the Hayes PROCESS because this method does the centring and interaction automatically and provides a more accurate and efficient test of moderation.
Chapter 4/ Study 3	Primary	Primary data selected from the 1000 Families study (see Appendix I). Respondents could either complete the	The aim of this study was to identify potential group differences in maternal outcomes. I made the decision to conduct

		survey online or request to have a hard copy posted to their home address.	ANOVAs because this is the most effective and efficient method of identifying group differences In addition, previous studies that have addressed the same research question have adopted this approach, thus enabling the results to be directly comparable. ANCOVAS effectively control the effects of other continuous variables providing the number of covariates does not exceed (.1 x sample size) – (the number of groups - 1). The number of covariates (n=5) in chapter 4 did not exceed the ideal amount therefore I decided on this analytical approach. Testing for effect sizes allowed me to identify existing group differences even when they could not be statistically identified via ANCOVAS. Therefore, I believed including this method would provide the most thorough insight into the results.
Chapter 5/ Study 4	Primary and Secondary	Primary and secondary data were used for this study. Primary data from the 1000 Families study (see Appendix I) was utilised. Respondents could either complete the survey online or request to have a hard	Study 4 investigated the measurement invariance of The Positive Gains Scale across three distinct groups. The two common approaches to test for measurement invariance are, comparing differences

		copy posted to their home address. This study also utilised secondary data from two sources. TD and CPHP data was obtained with permission from Dr Ineke Pit-ten Cate's doctoral research. Further secondary data were obtained with the permission Prof. Hastings who had conducted a UK wide study of families that have a child with autism. Petalas, M. A., Hastings, R. P., Nash, S., Hall, L. M., Joannidi, H., & Dowey, A. (2012). Psychological adjustment and sibling relationships in siblings of children with autism spectrum disorders: Environmental stressors and the broad autism phenotype. *Research in Autism Spectrum Disorders*, *6*, 546-555.	of χ^2 and more recently, comparing the differences in comparative fit index (ΔCFI). I decided to use both methods as there is some evidence that differences in χ^2 are sensitive to sample size. I believed that using both methods would provide more stringent results.