Clinical Psychology Review 110 (2024) 102426

Contents lists available at ScienceDirect

Clinical Psychology Review

journal homepage: www.elsevier.com/locate/clinpsychrev

Review

Psychological wellbeing in parents of children with Down syndrome: A systematic review and meta-analysis

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ARTICLE INFO

Keywords:
Down syndrome
Parents
Psychological wellbeing
Parenting stress
Parenting reward
Depressive symptoms

ABSTRACT

We report a review examining the psychological wellbeing of parents of children with Down syndrome (DS) relative to that of parents of typically developing (TD) children. A systematic search identified 57 relevant studies, which were synthesised meta-analytically. Relative to their counterparts with TD children, mothers and fathers of children with DS reported higher levels of parenting stress (mothers: $g = 0.57$, 95% CI [0.33, 0.81]; fathers: $g = 0.40$, [0.24, 0.56]), depressive symptoms (mothers: $g = 0.42$, [0.23, 0.61]; fathers: $g = 0.25$, [0.02, 0.48]) and psychological distress (mothers: $g = 0.45$, [0.30, 0.60]; fathers: $g = 0.63$, [0.26, 0.99]). Small effects were found for anxiety for mothers ($g = 0.16$, [0.03, 0.29]), with no differences for fathers ($g = 0.03$, [−0.25, 0.32]). No group differences were found for positive impact of parenting (mothers: $g = -0.09$, [−0.25, 0.07]; fathers: $g = -0.04$, [−0.30, 0.22]), while evidence concerning other positive wellbeing outcomes was limited. No significant moderating effects of child age range, country income level, or group differences in parental education level were identified, but limited subgroup analyses were possible. Raising a child with DS may be associated with elevated stress, depressive symptoms, and psychological distress for mothers and fathers. However, levels of parenting reward appear equivalent to those experienced by parents raising TD children.

Down syndrome (DS), also known as trisomy 21, is a genetic condition resulting from the presence of three copies of the 21st chromosome (Zhu et al., 2013). Like all children, those with DS have a wide range of abilities and characteristics; understanding the condition is not equivalent to understanding any individual child. DS is associated with intellectual disabilities which vary in degree, but are typically mild or moderate (Määttä et al., 2006). Developmental milestones may be reached at a different pace to children without disabilities, but learning continues into adulthood (Fidler & Nadel, 2007; Grieco et al., 2015). Children with DS can be meaningfully included in mainstream schools with individualised support and adaptations to optimise their learning (Faragher et al., 2020). DS involves increased susceptibility to certain medical conditions such as cardiac and gastrointestinal disorders (Bull, 2011), though medical advances have dramatically improved life expectancy in recent decades (Bittles et al., 2006). DS falls under the umbrella of developmental disabilities (DDs), a term encompassing varying conditions of childhood onset that affect motor, cognitive or social development (Odom et al., 2007).

Unlike many other developmental disabilities, DS can be identified prenatally through genetic testing. Recent decades have seen the expansion of available prenatal screening technologies, which has been reflected in changes to the DS prenatal testing pathway in many countries. Concurrently, disability rights scholars have expressed concern about the propensity for prenatal testing to perpetuate a reductive and negative view of DS (Owen et al., 2020; Parens & Asch, 1999). The realisation of informed choice with respect to prenatal testing depends upon the availability of relevant, balanced, and accurate information. Research indicates that the prospect of raising a child with DS is an important consideration for expectant parents when making decisions about prenatal testing and pregnancy continuation (Choi et al., 2012; France et al., 2012; Lawson, 2006). Healthcare professionals have a key role in supporting such decisions (Korenromp et al., 2007; Reed & Berrier, 2017) and hence require access to information about family experiences of DS that can be shared with prospective parents. While no

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https://doi.org/10.1016/j.cpr.2024.102426
Received 8 December 2022; Received in revised form 6 March 2024; Accepted 4 April 2024
Available online 6 April 2024

prenatal test can offer certainty about a family's future, the availability of clear evidence has an important role in permitting parents to make decisions aligned with their values and not unduly shaped by societal representations of DS.

Research addressing the wellbeing of parents raising a child with a DD has historically been aligned with narratives of family grief, crisis, or dysfunction (Ferguson, 2002). A substantial body of research documents higher levels of psychological distress among parents of children with DDs relative to those without disabilities. For example, previous reviews have found that parents of children with DDs have higher levels of depressive symptoms (Scherer et al., 2019; Singer, 2006) and stress (Lee, 2013; Masefield et al., 2020).

While such reviews have typically grouped parents of children with DS and other DDs together, evidence suggests that there may be substantive differences in the psychological wellbeing of these populations. The former are often reported to demonstrate lower parenting stress and higher parenting reward, in a phenomenon denoted the 'Down syndrome advantage' (Hodapp et al., 2001). Differences in child behavioural characteristics and in parental socio-economic status (SES) have each been implicated in the purported 'advantage' (Jess et al., 2021; Stoneman, 2007). Children with DS are often found to have lower rates of behaviour problems than children with other DDs such as autism (Einfeld et al., 2006). Parents of children with DS also tend to be older and may be socio-economically advantaged relative to parents of children with other DDs (Hodapp et al., 2012).

Research concerning the psychological wellbeing of parents of children with DS relative to parents of typically developing (TD) children has produced mixed findings. Some studies have reported greater stress or depressive symptomatology in the former group (Hedov et al., 2002; Phillips et al., 2017; Sanders & Morgan, 1997), while others have found no statistically significant group differences (Pastor-Cerezuela et al., 2021; Van Riper et al., 1992). The diversity of findings concerning this comparison indicates the need for a comprehensive synthesis of this body of research.

Recently, greater emphasis has been placed on how parents cope with stress, and develop and maintain resilience (McConnell & Savage, 2015), and on their experiences of positive outcomes related to raising a child with DD (Blacher, Baker, & Berkovits, 2013; Hastings, 2016). In a study focused on parents of children with DS, a large majority reported their child to be a source of love, pride, and a more positive outlook on life (Skotko et al., 2011). Increasing attention to positive elements of parental wellbeing aligns with the positive psychology movement which highlights that the absence of symptoms of mental ill health is not equivalent to psychological wellbeing (Iasiello et al., 2020; Kramer, 1997). Similarly, the presence of parenting stress does not preclude parenting rewards or positive outcomes (Beighton & Wills, 2019), as is frequently affirmed by participants in qualitative research (How et al., 2019; Korkow-Moradi et al., 2017; Pillay et al., 2012). Positive and negative impact are often considered to be orthogonal rather than opposite constructs (Horsley & Oliver, 2013). The importance of attending to both such facets in parents raising children with DDs is increasingly acknowledged (Cuskelly et al., 2009; Hastings, 2016).

Information about parental wellbeing in families raising a child with DS is of relevance to expectant parents navigating the DS prenatal screening pathway, and the healthcare professionals supporting them. However, evidence from studies comparing psychological wellbeing in parents of children with DS and TD has been mixed. Previous comparative reviews have focused on specific, usually negative, aspects of parental wellbeing such as parenting stress or depressive symptoms, and have tended to group parents of children with DS together with parents of children with other DDs. While the experiences of families of children with DS are diverse, they may share characteristics which warrant consideration of their experiences distinctly from those of other families. A comprehensive account of parental experiences includes consideration of both positive and negative aspects of psychological wellbeing. The aim of the present systematic review was to identify and synthesise existing evidence concerning a range of psychological wellbeing outcomes for parents of children with DS in comparison to parents of TD children.

1. Methods

The protocol for this review was published with PROSPERO (registration number CRD42021242521). The published protocol describes a wider review conducted as part of a PhD programme. The method and findings reported here correspond to the comparison between parents of children with DS and parents of non-disabled children.

1.1. Search strategy

The search strategy was designed to identify studies reporting quantitative data pertaining to positive and/or negative domains of psychological wellbeing in parents of children with DS and TD. The focus was on individual rather than family-level outcomes, including both those specific to parenting (i.e., parenting stress and parenting reward) and non-specific (e.g., depression, life satisfaction).

The search strategy comprised three elements:

1. The databases PsycINFO, Medline and Embase (OVID), Web of Science, (Clarivate) CINAHL (EBSCO), and ASSIA (ProQuest) were searched on 2nd March 2021, with a search update conducted on 12th September 2023.
2. The following websites were searched on 4th March 2021 and 13th September 2023: Down's Syndrome Research Foundation (UK and Canada), Trisomy 21 Research Society, Down's Syndrome Association (UK), Global Down Syndrome Foundation, Down Syndrome Education International.
3. Forward and backward citation searching of included studies was conducted using the Web of Science citation index.

Three search strings, respectively pertaining to DS, parents, and psychological wellbeing outcomes, were combined using the Boolean 'and' operator. No date limits were applied and (where possible) searches were limited to articles published in English and involving humans. The full database search strategy is shown in Appendix A.

1.2. Eligibility criteria

Studies were included which reported quantitative data about: (1) the psychological wellbeing of (2) parents of children (under-18 s) with DS, (3) compared to parents of children (under-18 s) without disabilities.

Studies were excluded if they were not reported in English, if the mean age of children was ≥18 years, if they did not report quantitative data from primary research (e.g., reviews and conference abstracts were excluded), or where >30% of the study sample did not meet the inclusion criteria.

1.3. Study selection process

The open-access online platform CADIMA (Kohl et al., 2018) was used to manage study selection. Two reviewers (TR and CM) independently screened abstracts, and assessed full texts of remaining reports, against the eligibility criteria. Disagreements were resolved through discussion, consulting a third reviewer (CS or RH) where necessary.

After full text assessment, study details were checked to identify potential duplicate or overlapping studies having been reported in multiple publications. Such publications were each retained if they provided differing data, but not included together in a single analysis.

1.4. Data extraction

A single reviewer (TR) extracted data from each report, with a second reviewer (CM) verifying the accuracy of key outcome data (sample sizes and summary statistics for each group on measures of psychological wellbeing). Data were also extracted for study design, country, instrument(s) used, participants' gender, ages of their children, indicators of SES, and measures of child behaviour problems (where available). Study authors were contacted for relevant data if unavailable in reports.

1.5. Quality appraisal

Quality appraisal was conducted independently by two reviewers (TR and CM), with disagreements resolved through discussion and in consultation with a third reviewer (CS) where necessary. Studies' quality was appraised using adapted critical appraisal checklists for cross-sectional and cohort studies from the Joanna Briggs Institute (Moola et al., 2020), details of which are available in Appendix D. Sensitivity analyses were conducted by removing studies meeting fewer than 50% of quality criteria to test for the influence of studies rated as lower in quality.

1.6. Effect measures

For continuous outcomes, standardised mean differences (SMDs) and their 95% confidence intervals (95% CI) were calculated using the formula for Hedges' *g* (Hedges & Olkin, 1985). Where studies reported data from multiple timepoints, data from the earliest timepoint for which suitable data were available were used to calculate effect sizes for inclusion in meta-analyses. This strategy was chosen in the interests of consistency and agreed prior to analysis, with sensitivity analyses conducted to check the impact of this approach.

1.7. Synthesis methods

Data from studies were grouped by the psychological wellbeing outcome they measured, with separate syntheses conducted for each. Parenting-specific outcomes (parenting stress and positive impacts from parenting) were distinguished from outcomes which are not specific to parenting experiences (e.g., depressive symptoms, psychological distress). The psychological distress analysis comprised data collected using measures of various psychiatric symptoms, which included symptoms both of anxiety and depression, rather than being focused on either one. Studies included in the life satisfaction analysis specifically measured an individual's personal cognitive evaluation of their life, while studies in the quality of life (QoL) analysis used broader, multidimensional measures targeting an "individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns" (WHOQOL Group, 1998, p.551). Appendix E contains further details of outcome classification.

For each outcome, separate syntheses of data from mothers and fathers were conducted, in recognition of potential gender differences in parental wellbeing (Dunn et al., 2019; Hauser-Cram et al., 2001).

Where there were sufficient data, planned subgroup analyses were conducted to investigate the impact of factors whose relationship with psychological wellbeing in parents of children with DDs have been most robustly demonstrated in the literature: parental socio-economic position (Emerson & Hatton, 2009) and child behaviour problems (Baker et al., 2002; Biswas et al., 2015). Child age has also been shown to correspond to psychological wellbeing in parents of children with DDs including DS (Most et al., 2006; Woodman, 2014). Because child age range can vary across studies, planned analyses were also conducted, where data allowed, to investigate the impact of this. For this purpose, studies were categorised as: those focusing only on parents of young children vs. those also including parents of older children; those conducted in high vs. middle income countries according to World Bank Group classifications; those reporting the presence vs. absence of statistically significant group differences in parental education level; and those reporting the presence vs. absence of statistically significant group differences in child behaviour problems.

When standard deviations were not reported and could not be obtained through contact with authors, these were imputed from another study in the review utilising the same instrument (in the event of multiple candidates, the study with the most comparable context and sample characteristics was selected). Studies reporting relevant outcome data which were not in the form of mean scores (e.g., median scores or dichotomous data) were included in the review but not in the meta-analyses; their findings were instead synthesised narratively.

For each psychological wellbeing outcome with appropriate data, a random-effects meta-analysis was conducted using Stata version 17.0 (StataCorp, 2021) to calculate a pooled effect size and 95% CI. A random-effects model was chosen in anticipation of between-study heterogeneity. Studies were weighted according to the inverse of the total variance, with restricted maximum likelihood estimation (REML) of the between-study variance. The robustness of meta-analytic findings to the influence of individual studies was investigated through leave-one-out analysis (Anzures-Cabrera & Higgins, 2010). Sensitivity analyses were conducted to investigate the impact of differing study designs and analytic strategies upon the pooled effect estimates and between-study heterogeneity.

Cochran's Q test was used to assess the presence of between-study heterogeneity. The proportion of total variance attributable to between-study heterogeneity was quantified using the I^2 statistic (Higgins et al., 2003) Where the presence of considerable between-study heterogeneity was evident, the influence upon heterogeneity of potential outliers and of predefined subgroups was investigated.

For meta-analyses including at least 10 studies (Page, Sterne, et al., 2021), Egger's regression test (Egger et al., 1997) and funnel plot analysis (Sterne et al., 2011) were used to assess the possible presence of small study effects.

2. Results

Fig. 1 displays the results of the study selection process. Appendix B details studies excluded at the full text stage, with the primary reason for each exclusion.

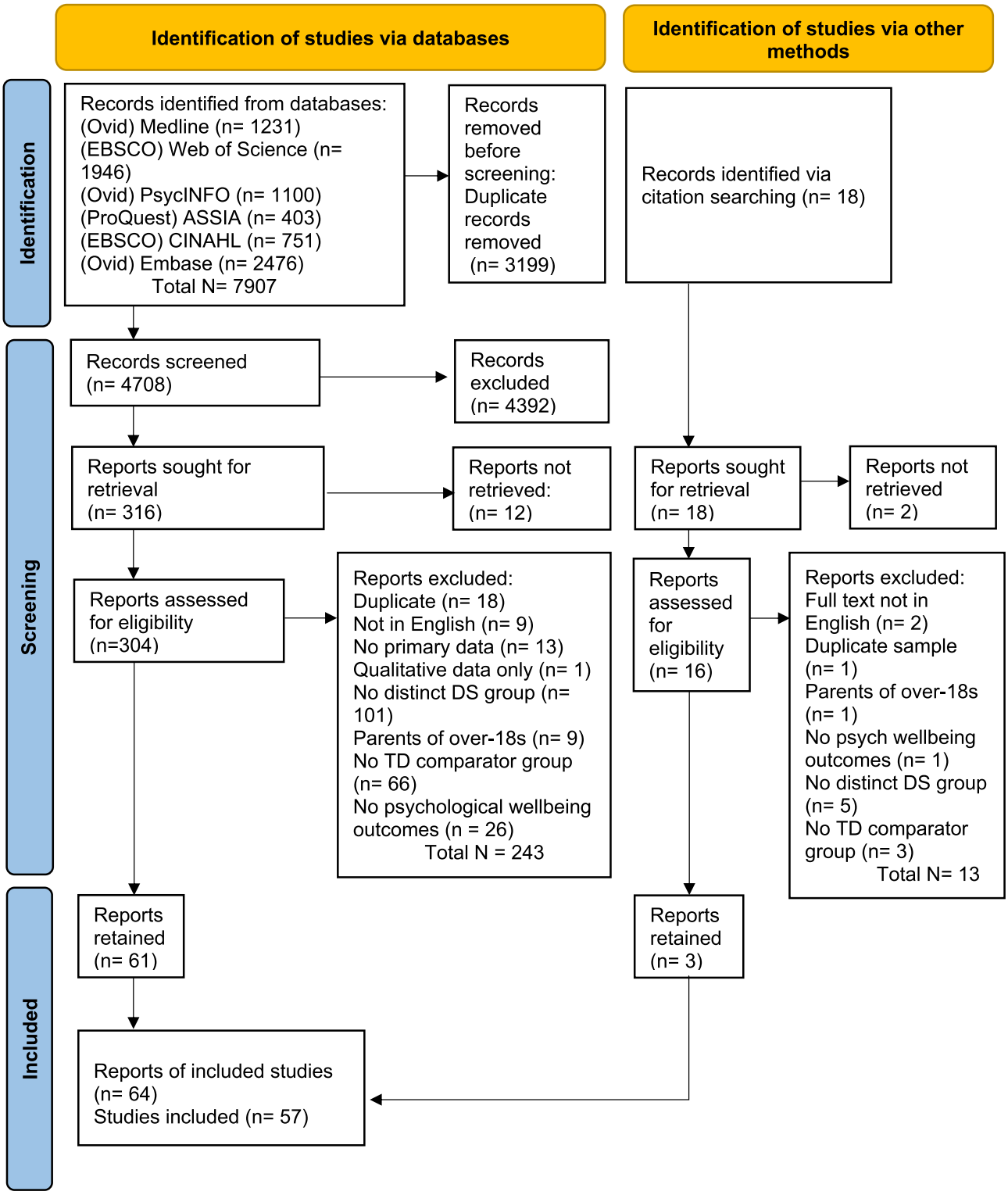
2.1. Study characteristics

Characteristics of included studies and a full reference list are shown in Appendix C. Fifty-seven studies (reported in 64 papers) were included in the final review, reporting data from a total of 383,796 parents. This latter number reflects the inclusion of two large population studies (Fairthorne et al., 2015; Nes et al., 2014). Seven studies were longitudinal while the remainder were cross-sectional in design.

The most frequently examined wellbeing outcomes were parenting stress, parenting distress or parenting burden (36 studies), depressive symptoms (20 studies), parenting reward, satisfaction, or positive impact (14 studies), psychological distress (10 studies), anxiety (8 studies), and quality of life (8 studies).

One study included only data from fathers (Rodrigue et al., 1992), 21 reported data only from mothers, and in a further 14 studies at least 75% of participants were mothers.

Forty-eight studies were conducted in high-income countries, of which 15 were conducted in the USA; four in Canada; three in Australia and the remainder in Europe. Nine studies were conducted in upper-middle or lower-middle income countries: Albania (Aliaj, 2017), Brazil (Barros et al., 2017, 2019; Dias et al., 2022), Jordan (Amireh, 2019); Turkey (Muammer et al., 2013; Senses Dinc et al., 2019); Pakistan (Fatima & Suhail, 2010); Poland (Pisula, 1998); and South Africa (Molteno & Lachman, 1996).



DS: Down Syndrome
TD: Typical development

Fig. 1. PRISMA diagram showing study selection process Page et al., 2021.

2.2. Quality appraisal

Results of quality appraisal are shown in Appendix D. There was a wide variety of quality appraisal ratings across studies, with some meeting only one or two of seven criteria, and several others meeting all of them. Studies generally performed well with respect to description of the research context and sample (81% of studies) and measurement of the outcomes using instruments that had been empirically tested and validated (86% of studies). The longitudinal studies also performed well in terms of the two groups deriving from similar populations (86% of studies). The poorest performance was in measurement of the 'condition' – that is, allocation of parents to the study and control groups (54% of studies). Those which did not meet this criterion often did not describe measures to ensure that parents in the TD group did not have children with DD. Among the longitudinal studies, only one (Nes et al., 2014) of three studies with incomplete follow-up reported strategies to address attrition. Another area of relative weakness was in the use of strategies to identify and address potential confounding, which appeared adequate in only 69% and 63% of studies respectively. Many did not report matching on the basis of socio-demographic variables, nor testing or taking steps to account for any such group differences.

3. Analysis

3.1. Parenting stress

3.1.1. Mothers

Twenty-one studies examined parenting stress in samples composed of at least 80% mothers. Parenting stress was higher in mothers of children with DS than mothers of TD children ($g = 0.57$, 95% CI [0.33, 0.81], $p < .001$; Fig. 2), but with considerable heterogeneity ($I^2 = 88\%$). Sensitivity analyses are detailed in Appendix F1. These identified that removal of the outlying study by Barros et al. (2019) reduced heterogeneity (to $I^2 = 67\%$), with its exclusion also reducing the SMD to 0.48 (95% CI [0.33, 0.63], $p < .001$), while removal of two other outlying studies (Amireh, 2019; Pastor-Cerezuela et al., 2021) appeared to have little impact on the findings. Removing three studies (Amireh, 2019; Gugliandolo et al., 2022; Phillips et al., 2017) which met fewer than 50% of quality criteria slightly increased the effect size ($g = 0.62$, 95% CI [0.36, 0.88], $p < .001$) but did not reduce heterogeneity ($I^2 = 89\%$). There was little impact of removing all studies whereby parent domain scores of stress scales were used (due to missing total scores; $g = 0.52$, 95% CI [0.29, 0.75], $p < .001$; $I^2 = 90\%$) or of removing the only study of longitudinal design (Eisenhower et al., 2005; $g = 0.59$; 95% CI [0.34, 0.83], $p < .001$; $I^2 = 89\%$). Leave-one-out analysis identified no highly influential studies (see Appendix F1).

There were no statistically significant subgroup differences in relation to child age range ($p = .19$), parental education level ($p = .25$), or country income level ($p = .77$); Appendix F1. Child age range did appear to impact heterogeneity, since there was no evidence of statistical heterogeneity between studies including parents of young children only (Q(3) = 1.51, $p = .68$; $I^2 = 0\%$).

Egger's regression test did not find evidence of small study effects ($z = -0.47$, $p = .64$), though a funnel plot showed some evidence of asymmetry, suggesting that publication bias may be impacting the findings (Appendix G1).

3.1.2. Fathers

Nine studies examined parenting stress in fathers. Parenting stress was higher in fathers of children with DS than fathers of TD children ($g = 0.40$; 95% CI [0.24, 0.56], $p < .001$; Fig. 2), with no evidence of heterogeneity (Q(8) = 9.01, $p = .34$; $I^2 = 0\%$). Leave-one-out analysis identified no highly influential studies (Appendix F2). Removing two studies (Amireh, 2019; Gugliandolo et al., 2022) which met fewer than 50% of the quality criteria had no substantive impact on the findings ($g = 0.40$, 95% CI [0.23, 0.57], $p < .001$). Subgroup analyses and tests of small study effects were not possible due to the small number of studies included.

3.1.3. Additional studies

Findings from 13 studies which were not included in the meta-analyses - due to non-availability of necessary data (9 studies) or because mothers' and fathers' data were not reported distinctly (4 studies) - are detailed in Appendix H. In brief, the majority were in keeping with the results of the meta-analyses, suggesting higher levels of stress in parents of children with DS than parents of TD children.

3.2. Depressive symptoms

3.2.1. Mothers

Fifteen studies examined depressive symptoms in mothers only, or in samples comprising at least 80% mothers. Depressive symptomatology was higher in mothers of children with DS than of children who are TD ($g = 0.42$, 95% CI [0.23, 0.61], $p < .001$; Fig. 3), though there was substantial heterogeneity between studies ($I^2 = 72\%$). Removal of an outlying study (Pastor-Cerezuela et al., 2021) resulted in a reduction in I^2 to 55% and a small reduction in the effect size ($g = 0.36$, 95% CI [0.20, 0.51], $p < .001$; Appendix F3). There were no substantial changes in heterogeneity or effect size estimates as a result of removing the only longitudinal study (Eisenhower et al., 2005; $g = 0.45$; 95% CI [0.27, 0.64], $p < .001$; $I^2 = 70\%$) nor as a result of removing the only study meeting fewer than 50% of the quality criteria (Muammer et al., 2013; $g = 0.42$, 95% CI [0.22, 0.62], $p < .001$; $I^2 = 70\%$; Appendix I1.1). Leave-one-out analysis indicates that these results are also robust to the removal of any other single study (Appendix F3).

There were no statistically significant subgroup differences in relation to child age range ($p = .25$) or group differences in parental education level ($p = .17$); Appendix F3. No further subgroup analyses were possible. Egger's regression test found no evidence of small study effects ($z = 0.51$, $p = .61$) and a funnel plot showed no evidence of asymmetry (Appendix G2).

3.2.2. Fathers

The meta-analysis of differences in depressive symptoms among fathers of children with DS and TD found a marginally statistically significant difference between the two groups ($g = 0.25$, 95% CI [0.02, 0.48], $p = .04$; Fig. 3), with evidence of moderate between-study heterogeneity (Q(6) = 12.17, $p = .06$; $I^2 = 51\%$). None of these studies met fewer than 50% of the quality criteria. There were no apparent outliers and no studies of longitudinal design. Leave-one-out analysis demonstrated that these findings are not robust; removal of any of four studies would render the overall effect estimate statistically non-significant (Appendix F4).

There were no statistically significant subgroup differences in relation to child age range ($p = .32$) or differences in parental education level ($p = .72$); Appendix F4. Testing for small study effects was not possible due to the small number of studies included.

3.2.3. Additional studies

Five additional studies investigated depressive symptoms but could not be synthesised meta-analytically, due to reporting median rather than mean scores (Dias et al., 2022; Ljubičić et al., 2020, 2022; Senses Dinc et al., 2019) or reporting dichotomous data (Carr, 1988; Gath, 1977). In keeping with the above findings, each study reported depressive symptoms to be higher in parents of children with DS than TD, though group differences were not always statistically significant (further details in Appendix H).

3.3. Psychological distress

3.3.1. Mothers

Seven studies examined psychological distress in samples comprising

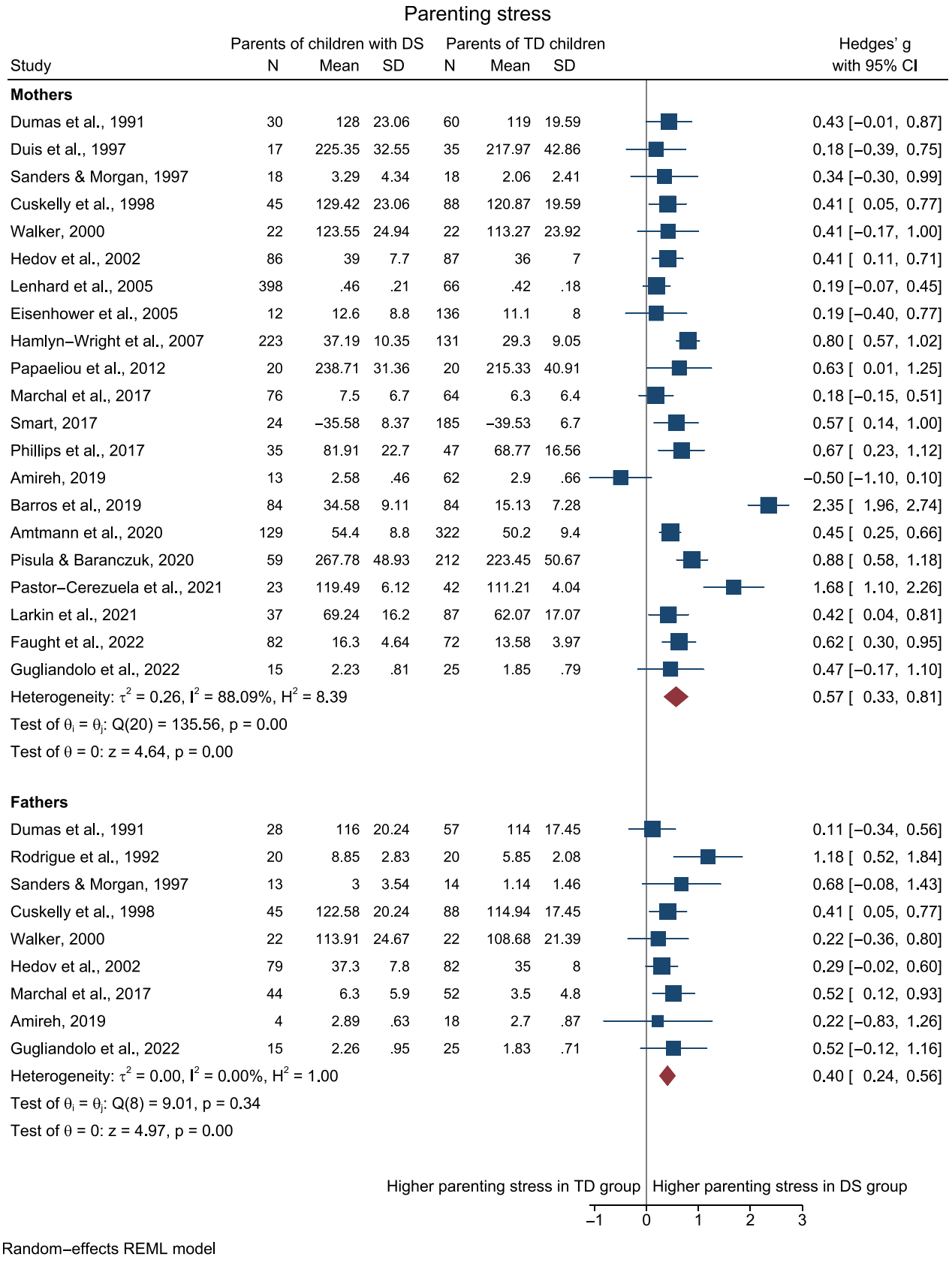


Fig. 2. Forest plots showing group differences in parenting stress for mothers and fathers.

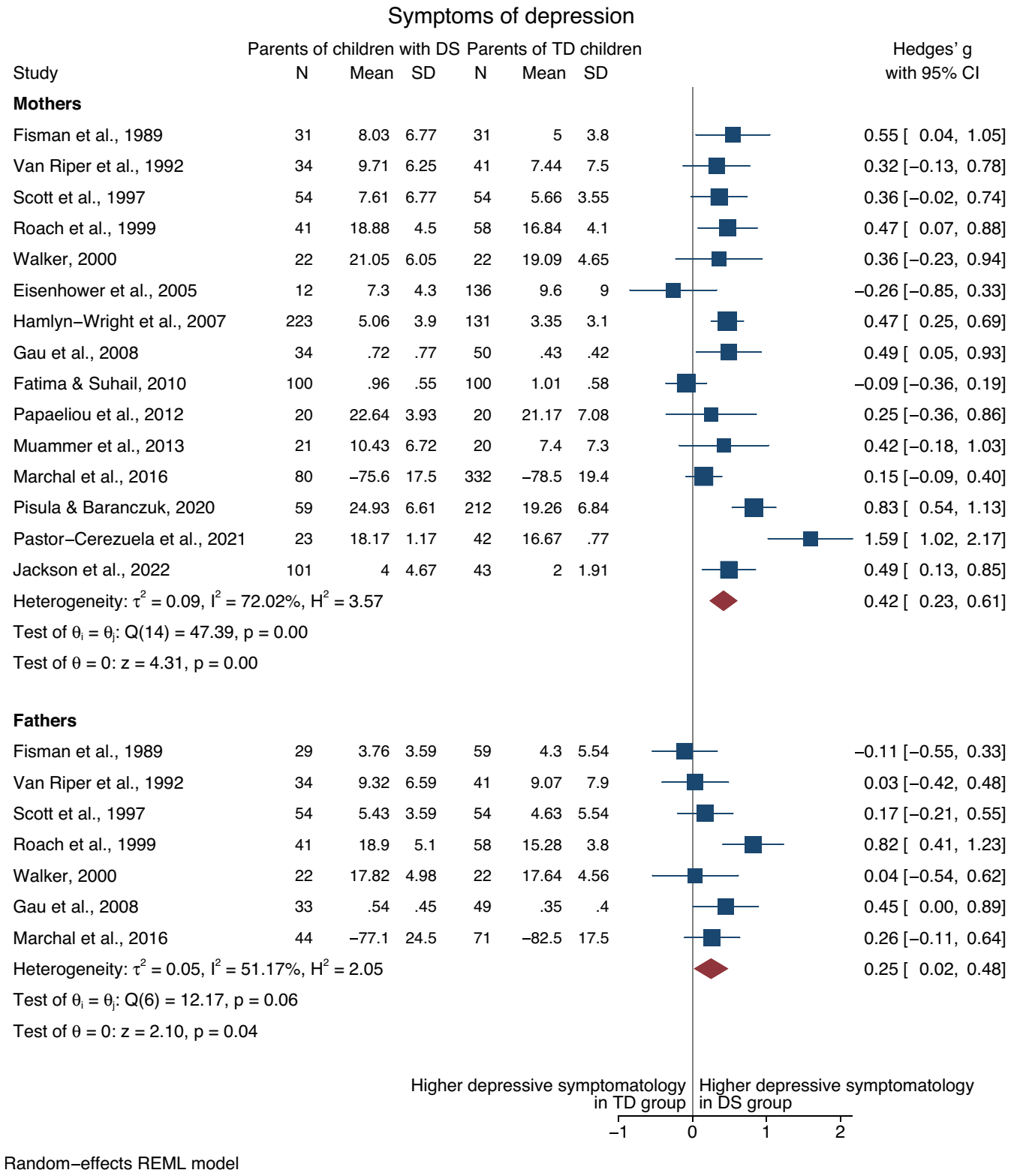


Fig. 3. Forest plots showing group differences in depressive symptomatology for mothers and fathers.

only mothers. Overall, mothers of children with DS reported higher levels of psychological distress than mothers of TD children (g = 0.45, 95% CI [0.30, 0.60], p < .001; Fig. 4). There was no statistical evidence of between-study heterogeneity (Q(6) = 6.50, p = .37; I^2 = 0%). Neither removing studies with imputed SDs (g = 0.44, 95% CI [0.14, 0.77], p = .004), nor removing studies meeting fewer than 50% of the quality criteria (g = 0.47, 95% CI [0.31, 0.62], p < .001), nor removing longitudinal studies (g = 0.43, 95% CI [0.18, 0.68], p < .001), had a substantive impact on the findings (Appendix F5). No subgroup analyses were possible. Leave-one-out analysis indicated that the findings are robust to the removal of any single study (Appendix F5).

Three of these studies also reported the proportion of mothers reaching clinical cut-off scores on psychological distress measures (see Appendix H). The largest of these (Nes et al., 2014) found statistically

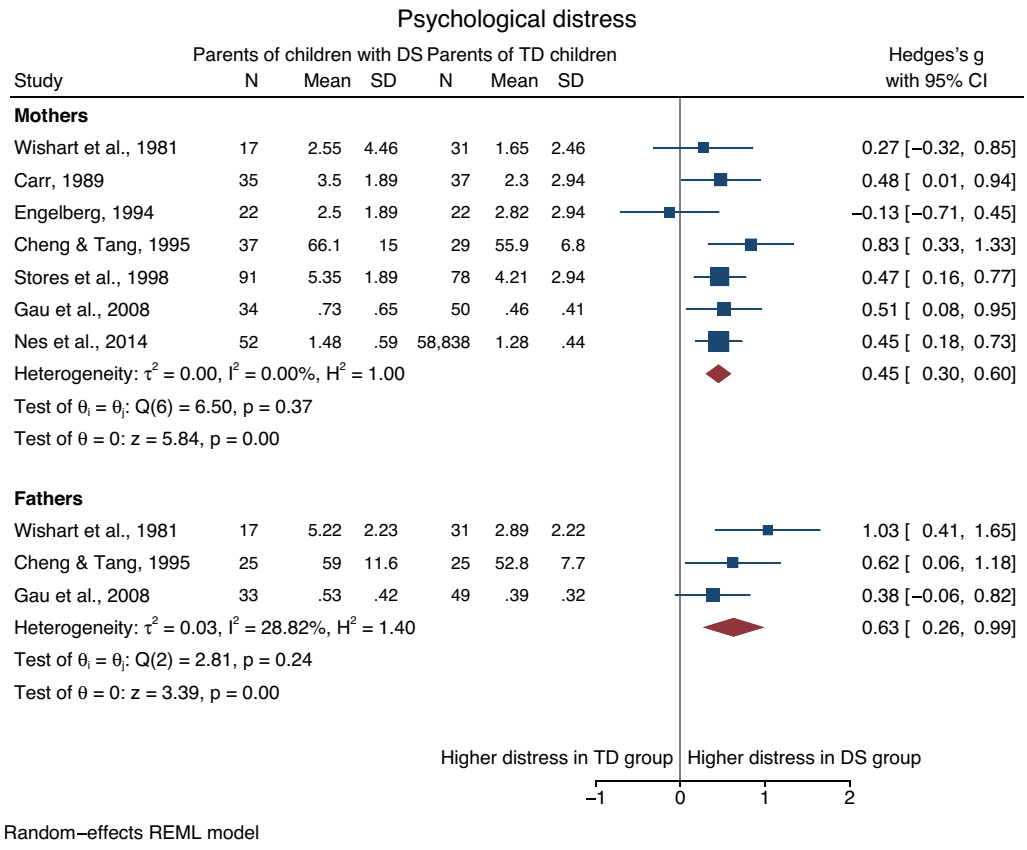


Fig. 4. Forest plots showing group differences in psychological distress for mothers and fathers.

significant group differences favouring the TD group when children were aged 6 months (OR 2.33, 95% CI [1.24, 4.40], $p = .01$) and 36 months (OR 3.12, 95% CI [1.6, 6.1], p = .01), but not at 18 months (OR 1.79, 95% CI [0.89, 3.60], $p = .08$). Differences were non-significant in the other two studies (Carr, 1988; Stores et al., 1998) with mothers of school-aged children.

3.3.2. Fathers

Three studies reported psychological distress data for fathers distinctly. Fathers of children with DS reported higher psychological distress than fathers of TD children (g = 0.63, 95% CI [0.26, 0.99], p < .001; Fig. 4), with no evidence of between-study heterogeneity (Q(2) = 2.81, $p = .24$; $I^2 = 28.82\%$). Removing the study whose SDs were imputed (Wishart et al., 1981) reduced the pooled effect size to 0.47 (95% CI [0.13, 0.82], $p = .007$; Appendix F6). Only one of these studies (Gau et al., 2008) met at least 50% of the quality criteria.

3.3.3. Additional studies

Three further studies, which each focused on parents of young children (Gath, 1977; Pelchat et al., 1999; Senses Dinc et al., 2019), reported psychological distress findings which were not amenable to inclusion in the above analyses. Their results largely align with those above, finding higher distress in parents of children with DS than those with TD children (Appendix H).

3.4. Anxiety symptoms

3.4.1. Mothers

Five studies reported data concerning symptoms of anxiety in mothers. Overall, there was a statistically significant difference of very small size, with mothers of children with DS reporting higher anxiety than mothers of TD children ($g = 0.16$, 95% CI [0.03, 0.29], $p = .01$; Fig. 5). There was no statistical evidence of between-study heterogeneity (Q(4) = 6.59, $p = .16$; $I^2 = 0\%$). None of these studies met fewer than 50% of the quality criteria. Leave-one-out analysis indicated that these findings are not robust; removal of any of three studies (Gau et al., 2008; Hamlyn-Wright et al., 2007; Lenhard et al., 2005) results in a non-significant overall group difference (Appendix F7).

There were no statistically significant subgroup differences in relation to child age range ($p = .29$; Appendix F7). Further subgroup analyses and tests of small study effects were not possible.

3.4.2. Fathers

Two studies reported data for fathers separately; overall there were no group differences in fathers' anxiety symptoms ($g = 0.03$, 95% CI [−0.25, 0.32], $p = .81$; Fig. 5) with no observed between-study heterogeneity (Q(1) = 0.12, $p = .73$; $I^2 = 0\%$).

3.4.3. Additional studies

Three studies (Dias et al., 2022; Ljubičić et al., 2020, 2022; Senses Dinc et al., 2019) could not be included in the above analyses since they reported median (rather than mean) anxiety scores. None of these studies found statistically significant group differences in anxiety between parents of children with DS and TD.

3.5. Incidence of mental health conditions

Three longitudinal studies examined the incidence of mental health conditions, measured via contact with healthcare services for psychological concerns, in parents of children with DS and TD (Appendix H2). The largest of these (Fairthorne et al., 2015) was a retrospective cohort study of mothers' health records. This reported an adjusted Incidence Rate Ratio for the DS group compared with the TD group of 0.82 (95% CI [0.4, 1.6], non-significant) for healthcare contact relating to any psychiatric condition. Studies by Gath (1977) and Murdoch and Ogston (1984) including both mothers and fathers also reported finding no

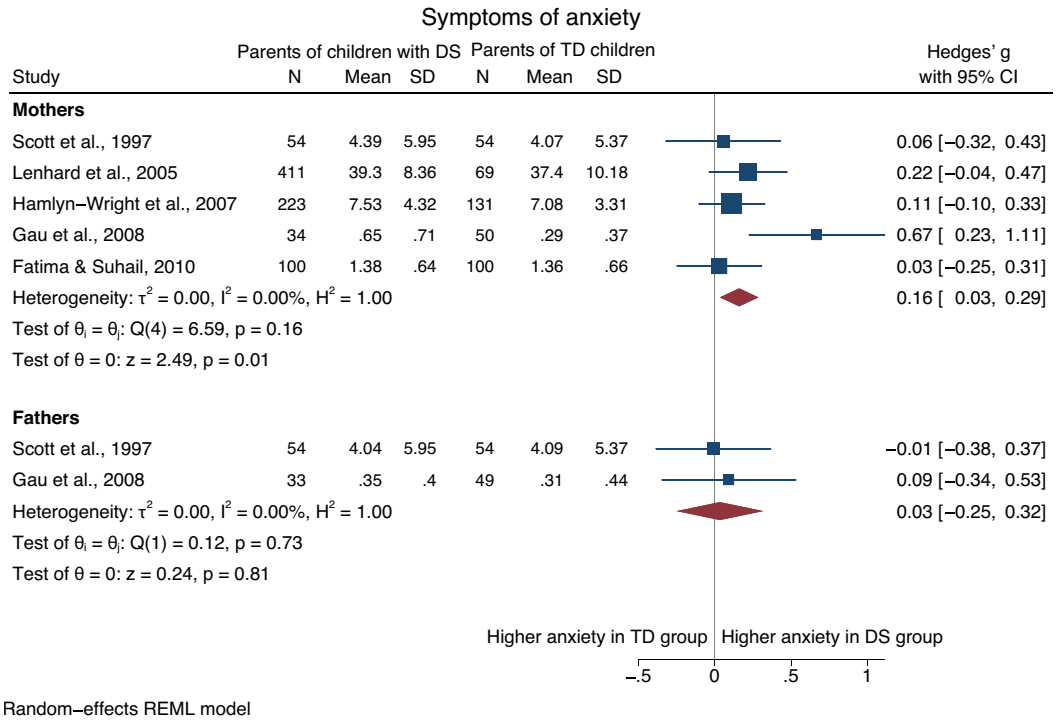


Fig. 5. Forest plots showing group differences in anxiety for mothers and fathers.

statistically significant group differences in the number of parents receiving healthcare contact for psychological problems in the years following the birth of their child.

3.6. Positive impact of parenting

3.6.1. Mothers

Eleven studies examined positive parenting outcomes in samples consisting of over two-thirds mothers. There was no statistically significant difference between groups of mothers of children with DS and TD ($g = -0.09$, 95% CI [−0.25, 0.07], $p = .25$; Fig. 6), and between-study heterogeneity did not appear substantial ($I^2 = 37\%$). Sensitivity analyses indicated the findings are robust to study design, quality rating, and analytical decisions, though leave-one-out analysis identified that omitting the study by Rodrigue et al. (1990) results in a very small, marginally statistically significant difference favouring the TD group (Appendix F8). There were no statistically significant subgroup differences in relation to child age range ($p = .67$; Appendix F8). No further subgroup analyses were possible.

Egger's regression test was statistically significant ($z = 2.74$, $p = .006$), suggesting that this analysis may be subject to small study effects. A funnel plot showed some evidence of asymmetry, suggesting that there may be studies missing from the review, especially those finding higher positive impact in parents of TD children (Appendix G3).

3.6.2. Fathers

Meta-analysis of four studies examining positive parenting outcomes in fathers found no statistically significant group difference ($g = -0.04$, 95% CI [−0.30, 0.22], $p = .78$; Fig. 6), with low between-study heterogeneity ($I^2 = 13\%$). Subgroup analyses and tests of small study effects were not possible. Removing one study which met fewer than 50% of the quality criteria (MacInnes, 2009) did not alter the overall finding of statistical insignificance ($g = -0.15$, 95% CI [−0.48, 0.18], $p = .38$; Appendix F9).

3.6.3. Additional studies

Three further studies of positive parenting impact were not included in the meta-analyses due to non-availability of compatible data (Appendix H). Briefly, in keeping with the above findings, two studies (Dabrowska & Pisula, 2010; Noh et al., 1989) found no statistically significant group differences in positive impact of parenting, while the third (Boström et al., 2010) did not report testing for such differences.

3.7. Life satisfaction and positive psychological wellbeing

Three studies reported life satisfaction scores in mothers of children with DS and TD. Overall, there was a small but statistically significant difference, with higher life satisfaction found in mothers of TD children than mothers of children with DS ($g = -0.33$, 95% CI [−0.57, −0.10], $p = .01$; Fig. 7). There was evidence of moderate between-study heterogeneity ($Q(2) = 2.90$, $p = .23$; $I^2 = 41\%$). One of these studies (Gugliandolo et al., 2022) met fewer than 50% of the quality criteria.

Gugliandolo et al. (2022) also examined life satisfaction in fathers, finding no statistically significant difference between fathers raising TD children and children with DS.

In one further study which examined the proportion of parents in each group rating themselves as satisfied with life (Branholm & Degerman, 1992), group differences were non-significant for both mothers and fathers.

Four studies examined various positive elements of psychological wellbeing besides life satisfaction; their findings are shown in Appendix H3. Each study reported no statistically significant group differences, though the TD group tended to score higher in each case. In one study (Marchal et al., 2016), group differences in favour of the TD group were significant at the 0.05 p-level, but not at the Bonferroni-adjusted level of 0.004.

3.8. Quality of life (QoL)

Three studies reported mothers' mean scores on the WHOQOL-BREF (WHOQOL Group, 1998). As shown in Fig. 8, group differences were not statistically significant with respect to overall perception of QoL ($g = -0.59$, 95% CI [−1.52, 0.34], $p = .21$), but psychological domain scores were higher in the TD group ($g = -0.60$, 95% CI [−1.09, −0.11], $p =$

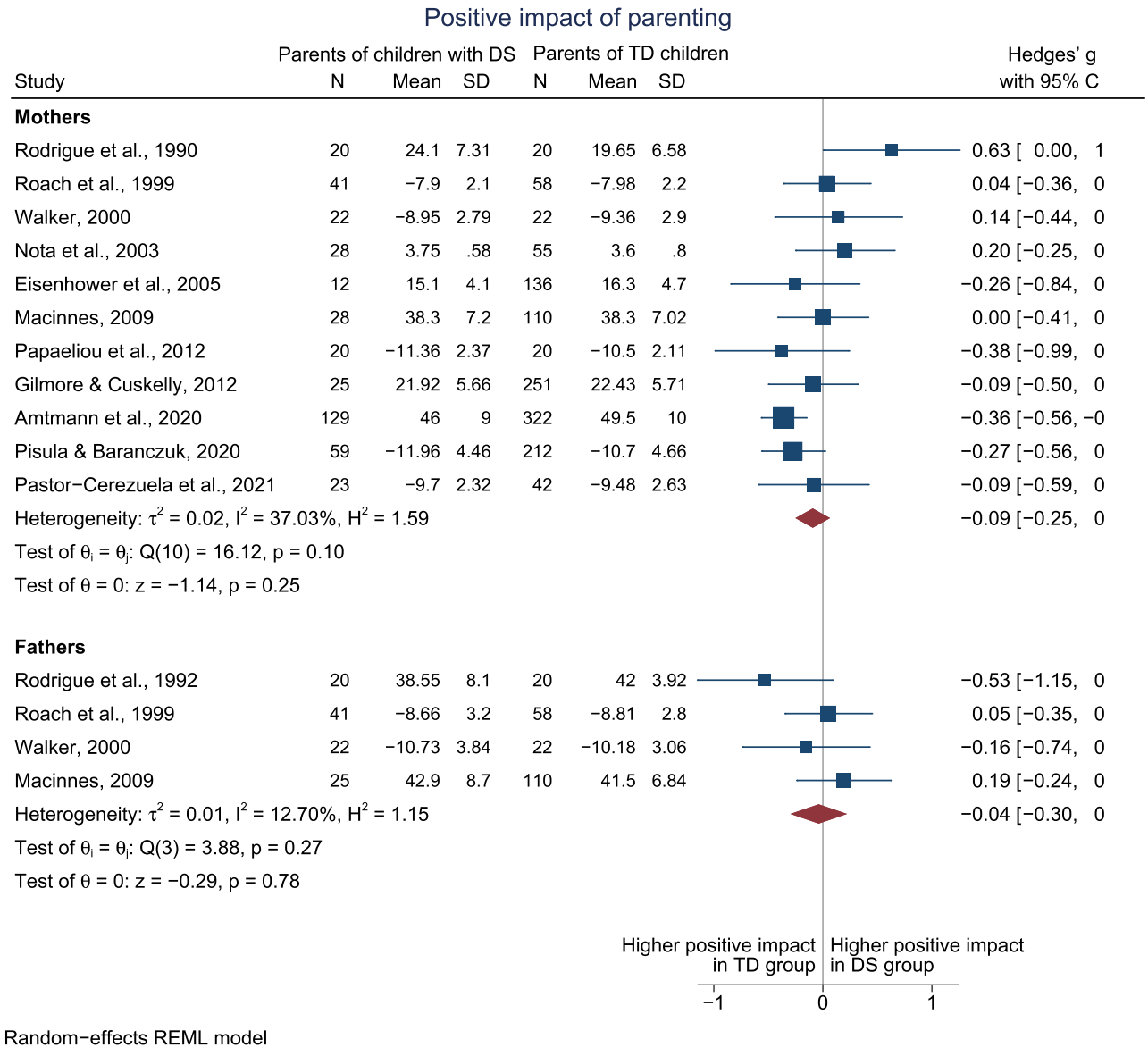


Fig. 6. Forest plots showing group differences in positive impact of parenting for mothers and fathers.

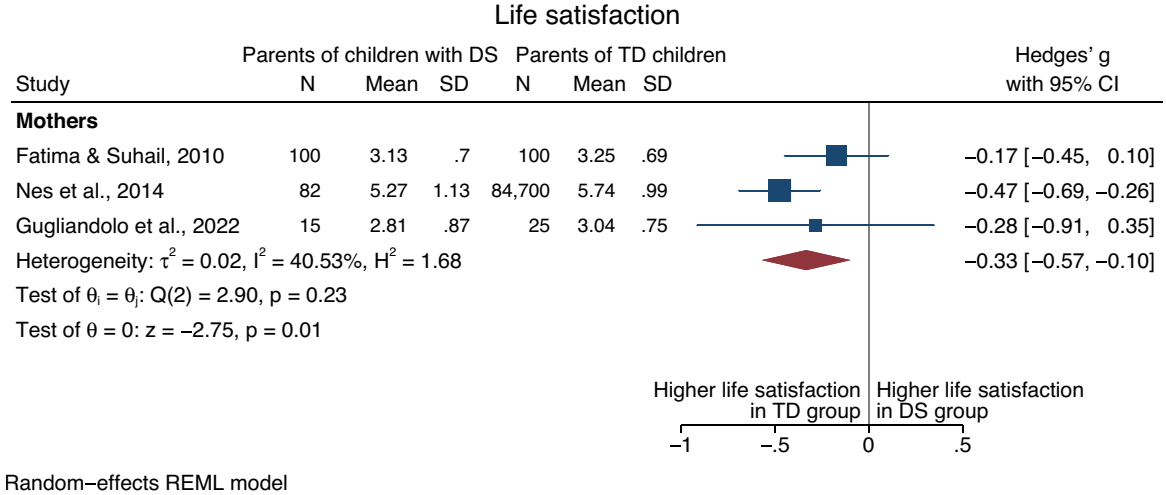


Fig. 7. Forest plot showing group differences in life satisfaction for mothers.

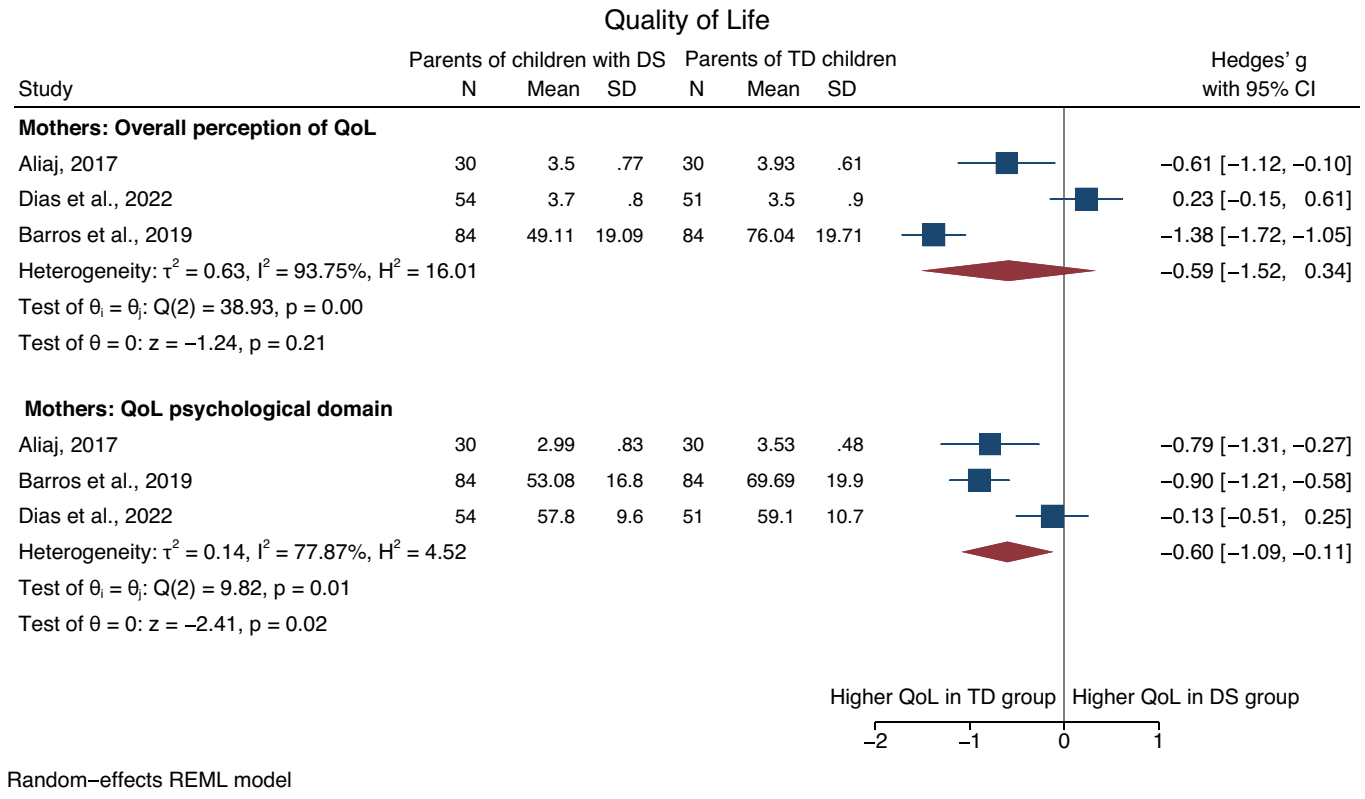


Fig. 8. Forest plots showing group differences in quality of life for mothers.

.02). Particularly in the former analysis, between-study heterogeneity appeared high ($Q(2) = 38.93$, $p < .01$; $I^2 = 94\%$). All studies derive from middle-income countries and two of the three (Aliaj, 2017; Dias et al., 2022) met fewer than 50% of the quality criteria.

Five further studies examined QoL, but their data were incompatible with the above meta-analyses and instead are shown in Appendix H. With respect to psychological domains of QoL, one of these studies (Hedov et al., 2000) reported that mothers and fathers of TD children scored significantly higher than their counterparts with children with DS. In three of the four remaining studies, group differences in psychological QoL domains were not statistically significant, though parents of TD children tended to score higher.

4. Discussion

This study reviewed evidence of differences in psychological wellbeing between parents of TD children and children with DS. We included a variety of outcomes concerning positive and negative domains of psychological wellbeing and examined group differences for mothers and fathers distinctly.

The main findings of this review can be summarised as follows. First, mothers and fathers of children with DS scored higher than their counterparts with TD children on measures of parenting stress, psychological distress, and depressive symptoms. Smaller differences in parenting stress and depressive symptoms were found between the groups of fathers than of mothers. Concerning symptoms of anxiety, group differences were negligible for mothers and statistically insignificant for fathers. The incidence of mental health conditions also did not differ between parental groups in the three studies which examined this.

Second, mothers and fathers of children with DS were not found to differ from those having TD children with respect to positive impacts of parenting. Studies concerning other positive aspects of psychological wellbeing were small in number and diverse in the constructs they examined. With respect to life satisfaction and psychological domains of QoL, mothers of TD children scored higher than mothers of children with DS. Evidence concerning these outcomes in fathers was very limited. There were no statistically significant group differences in individual studies examining other positive psychological wellbeing outcomes.

The present findings correspond in several ways to the existing literature concerning psychological wellbeing in parents of children with DDs. Despite the prominent notion of a 'Down syndrome advantage' in parental wellbeing relative to parents of children with other DDs (e.g., Hodapp, 2007), the current findings highlight that parents of children with DS may nevertheless experience elevated stress and depressive symptomatology relative to parents of TD children. This aligns the findings with previous reviews concerning parents of children with intellectual and developmental disabilities, who are consistently found to report higher stress and depressive symptoms than parents of TD children (Lee, 2013; Masefield et al., 2020; Scherer et al., 2019; Singer, 2006). The findings regarding anxiety are also concordant with those of Scherer et al., who did not find statistically significant differences between the anxiety levels of parents of TD children and those raising autistic children and children with cerebral palsy.

In literature concerning parents of children with DD, stress and psychological problems have been consistently associated with behavioural concerns and socio-economic position (e.g., Bailey et al., 2019; Emerson et al., 2006; Neece et al., 2012; Olsson & Hwang, 2008). Subgroup analyses corresponding to these prespecified factors were limited by data availability however, and cannot provide direct insights into the findings. While some studies indicate that children with DS may exhibit higher levels of behaviour problems than TD children (Van Gameren-Oosterom et al., 2011), in the present review only eight studies examined group differences in child behaviour problems. Of these, the two which found greater behaviour problems in children with DS than TD also reported parental wellbeing differences favouring the TD group (Gau et al., 2008; Phillips et al., 2017). The other six studies found group differences neither in child behaviour problems nor parental wellbeing (Dumas et al., 1991; Eisenhower et al., 2005; Kasari & Sigman, 1998; Ljubičić et al., 2020; Papaeliou et al., 2012; Stores et al., 1998). With

respect to socio-demographics, a number of studies in this review reported differences in some measure of SES favouring the TD group, with none reporting differences in the opposite direction. Although parents of children with DS may be of higher SES than parents whose children have other DDs (e.g., Stoneman, 2007) they may nevertheless be socio-economically disadvantaged relative to parents of TD children (e.g., Budd et al., 2015). Financial hardship may also increase because of costs associated with raising a child with disabilities (Grosse, 2010).

Although we did not test for gender effects in the current review, differences found in parenting stress and depressive symptoms between groups of mothers were larger than those between the groups of fathers. This appears consistent with a prior review by Dunn et al. (2019) reporting parenting stress and depressive symptoms to be higher in mothers than fathers of children with intellectual disability (ID). Despite shifts towards greater egalitarianism in parenting norms over recent decades (Preisner et al., 2020), there are likely to remain gendered factors involved in parental wellbeing. Studies indicate that the tendency for mothers to take on a greater share of caregiving responsibilities is enduring (Craig & Mullan, 2011; Musick et al., 2020), particularly where children have additional care needs (Vinck & Brekke, 2020). The rise of intensive parenting norms may be particularly impactful upon mothers (Nomaguchi & Milkie, 2020), increasing their susceptibility to parenting stress (Rizzo et al., 2013). Fathers, on the other hand, may have difficulty consolidating their wish for active involvement in parenting their child with DD and their necessary or culturally expected role as 'breadwinner' (Shorey & Pereira, 2023). Previous research has demonstrated that fathers of children with DD can experience elevated stress and depressive symptoms relative to fathers of TD children (Giallo et al., 2015; Langley et al., 2020), and the present findings highlight that this also applies to fathers of children with DS. With respect to psychological distress, differences among fathers were in fact more marked than those among mothers, though two of the three studies reporting fathers' psychological distress appeared to have considerable methodological limitations.

The finding that positive impacts of parenting did not differ between groups supports the notion that increased parenting stress does not inevitably correspond to reduced parenting rewards (Counselman-Carpenter, 2017; Marshak et al., 2019). This underlines the importance of attending to both these facets of the parenting experience. Some studies have found analogous results, whereby for parents of children with DD the degree of positive impact has not differed from that of parents raising TD children (Baker et al., 2002; Blacher, Begum, et al., 2013), though such evidence is limited. Previous research suggests a negative relationship between child behaviour problems and parental perceptions of positive impact (Blacher & Baker, 2007), but we were not able to explore this factor in the current review. On the other hand, the meta-analytic findings indicated life satisfaction and psychological QoL to be higher in mothers of TD children than those with a child with DS. It may be that parenting demands, such as time and financial pressures, can offset parenting rewards with respect to life satisfaction and other broader indices of psychological wellbeing (Pollmann-Schult, 2014). This 'demands-rewards perspective' (Nomaguchi & Milkie, 2020) might be particularly relevant to parents of children with DS, who are likely to face additional 'demands,' such as increased caregiving time (Miller et al., 2015), and the need to arrange and advocate for specialist healthcare and educational input for their child (McGuire et al., 2004). It is notable however that the overall effect sizes for life satisfaction and psychological QoL were small, and each meta-analysis included only three studies. Further individual studies found no statistically significant group differences in mood level (Fatima & Suhail, 2010), parental sense of wellbeing (Nota et al., 2003) or a six-dimensional measure of psychological wellbeing (Van Riper et al., 1992) (constructs whose diversity precluded meta-analytic synthesis). Overall, there is hence insufficient evidence to draw strong conclusions about comparative levels of positive psychological wellbeing in these groups.

4.1. Limitations and future research

In the meta-analyses of mothers' parenting stress and depressive symptoms, between-study heterogeneity appeared high and could not be fully accounted for. There was also some evidence of small study effects in the analyses of mothers' positive parenting impact and parenting stress, which might be attributable to the omission of eligible studies as a result of publication bias.

The operationalisation of positive parenting impact was varied, perhaps reflecting the diversity of positive outcomes in parental accounts (Beighton & Wills, 2019) and a lack of consensus about their conceptualisation (Jess et al., 2017). This may have contributed to between-study heterogeneity, and may serve to obscure other effects, such as those associated with child age. For example, positive impact in terms of parenting reward might reduce as children age (Hodapp et al., 2001), while positive 'personal transformations' might increase (Scorgie & Sobsey, 2000). Furthermore, some researchers have suggested that the positive impacts of raising children with disabilities might include 'special benefits' dissimilar to those found in parents raising TD children (Blacher, Baker, & Berkovits, 2013), perhaps making quantitative comparison of positive outcomes problematic. Future research might work towards increasing consistency in the conceptualisation and measurement of positive aspects of parenting in these groups.

Subgroup analyses identified no statistically significant moderating effects, but as discussed, these analyses were substantially limited by the available data, in turn limiting the review's ability to delineate factors associated with the wellbeing differences identified. Furthermore, there are a number of factors with established relevance to parental wellbeing which were not examined in this review, including parental social support and coping styles. These are likely to have important effects upon the relationship between parenting demands and psychological wellbeing outcomes (e.g., Halstead et al., 2017; Van Der Veek et al., 2009) and the current findings indicate the importance of future research in this area.

The current review included only studies published in the English language, with the majority (48 of 57) conducted in high-income countries; generalisability to low- and middle-income countries (LMIC) is hence unclear. Differing experiences of raising children with DS in LMIC may correspond, for example, to reduced availability of support services (Mkabile et al., 2021); and poorer healthcare and health outcomes (Zahari et al., 2019). Furthermore, country income level is not the only relevant cross-national factor. Societal attitudes to disability are likely to impact upon parental wellbeing (Huiracocha et al., 2017), while gender effects may vary in line with cultural expectations of parenting roles (Choi & Van Riper, 2017). This presents another possible limitation to generalisability of the current findings, the majority of which derive from northern America, Australia, and Europe.

The current findings also span a period of considerable expansion in the landscape of prenatal testing for DS (Wilmot et al., 2023). There are concerns that such developments can exacerbate stigma experienced by those with DS and their families (Nuffield Council on Bioethics, 2017), highlighting the condition as undesirable and invoking negative assumptions about the parenting experience (Lalvani, 2011). Mothers in particular may experience blame or disapproval for deviating from the expected course of prenatal testing and subsequent termination (Landsman, 2008; Lawson, 2003) especially as new technologies make this more accessible (Birko et al., 2018). The relationship of parental wellbeing to evolving prenatal testing strategies will be an important area of future study.

Finally, the current review was focused on individual parental wellbeing, and cannot attest to group differences in relation to family-level outcomes. Several studies report parents raising children with DS and TD not to differ with respect to marital satisfaction, family cohesion (Santamaria et al., 2012; Thomas & Olson, 1993) or divorce rates (Lederman et al., 2015; Urbano & Hodapp, 2007). Such outcomes are vital to understanding and supporting family wellbeing, but their

exploration was beyond the scope of the present work.

4.2. Implications

Elevated stress and other psychological problems may impact a parent's ability to optimally support their child's development (Conners-Burrow et al., 2014) and engage effectively in parenting interventions (Crnic et al., 2017). Practitioners supporting families with a child with DS might consider incorporating assessment of parental psychological wellbeing to identify parents in need of additional support. Such supports could include access to stress-reduction interventions; evidence indicates that psychological interventions such as mindfulness-based programmes and cognitive-behavioural therapies can help to reduce stress and depressive symptomatology in parents of children with DD (Chua & Shorey, 2022; Lindo et al., 2016; Sohmaran & Shorey, 2019). Behavioural parent training programmes may reduce child behaviour problems and enhance parental wellbeing, and may be most effective when implemented in conjunction with psychological strategies (Crnic et al., 2017; Singer et al., 2007). However, behaviour problems appear less commonly in children with DS than with other DDs (e.g., Griffith et al., 2010) and other sources of stress such as caring responsibilities (Hedov et al., 2002), medical concerns (Bourke et al., 2008) and difficulties accessing adequate healthcare and educational support (Minnes & Steiner, 2009; Van den Driessen Mareeuw et al., 2019) may be more salient to many parents. This might increase the relative importance of targeted supports including links to parent-led organisations, timely information provision (Skelton et al., 2021) and family-centred respite services (Collins et al., 2013). The current findings demonstrate the value of research into specific approaches to supporting the psychological wellbeing of parents of children with DS.

At the same time, the findings indicate the importance for researchers and practitioners of recognising that parents of children with DS have positive experiences to an extent that may well be comparable to that of other parents. In the context of prenatal testing for DS, prospective parents and their healthcare providers require clear and accurate information about the condition and what it might mean for families. While family experiences of DS are diverse and multi-faceted, evidence such as that presented here comprises one form of information that can be drawn upon to support decision-making. In efforts to present a balanced view of the condition, information about potential positive impacts of raising a child with DS should be available in addition to information about potential challenges.

For current parents, perceiving positive impacts from parenting may play an important role in reducing the effects of parenting stress and enhancing parental wellbeing (Hastings & Taunt, 2002; Trute et al., 2010). Parents of children with DS have identified a positive attitude as an important element of their coping (Nelson Goff et al., 2016). This illustrates the importance of making positive narratives available and supporting the development, application, and preservation of positive perceptions (Horsley & Oliver, 2013). Support from other parents within peer support schemes may help to develop and affirm parents' sense of competence and positive outlook (Shilling et al., 2013). Practitioners also have a vital role in conveying messages of reassurance and hope during clinical encounters (Clark et al., 2020; Docherty & Dimond, 2018). Since parents of children with DS have described the profound impact that communication about their child's diagnosis can have on their expectations and subsequent experiences (Nelson Goff et al., 2013; Pillay et al., 2012), exposure to positive narratives is relevant from the earliest stages of the parenting journey.

Role of funding sources

This research was funded by the Economic and Social Research Council (ESRC) and Down Syndrome UK as part of a doctoral training partnership award. ESRC Midlands Graduate School DTP award reference number: ES/P000771/1.

Contributors

T. L. Rutter: Methodology, Investigation, Analysis, Writing - Original Draft. R. P. Hastings: Conceptualisation, Methodology, Supervision, Writing – Review & Editing. C. A. Murray: Validation, Investigation, Writing – Review & Editing. N. Enoch: Conceptualisation, Writing – Review and Editing. S. Johnson: Methodology, Resources, Writing – Review & Editing. C. Stinton: Conceptualisation, Methodology, Investigation, Writing – Review & Editing, Supervision.

Declaration of competing interest

This review is associated with the PhD studentship awarded to T.R. and supervised by C.S. and R.H. The studentship has been partially funded by Down Syndrome UK, a family-led organisation supporting families of children with Down syndrome. N.E. is the founder and chief executive of Down Syndrome UK. The remaining authors have no conflicts of interest to declare.

Data availability

Data will be made available on request.

Acknowledgements

The authors are grateful to Dr. Nick Parsons for his advice concerning the analytic strategies used in this project.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.cpr.2024.102426.

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