Systematic Review

Mental ill-health in mothers of people with intellectual disabilities compared with mothers of typically developing people: a systematic review and meta-analysis

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Abstract

Background Mothers of people with intellectual disabilities (IDs) face exceptional challenges and may be more prone to experiencing mental ill-health compared with mothers of typically developing people. These mental ill-health problems may differ at different stages of the caregiving trajectory. However, there is no evidence synthesis on this topic. We aimed to systematically review evidence in this area and identify gaps in the existing literature.

Method Prospero registration: CRD42018088197. Medline, Embase, CINAHL and PsycINFO databases were searched. No time limits were applied. Studies were limited to English language. Inclusion criteria were studies of mothers of people with IDs that also included a comparison group of mothers of typically developing/developed children. Data were extracted from selected studies using a structured database. Study selection and quality appraisal were double rated. Where possible, meta-analyses were performed.

Results Of the retrieved articles, 32/3089 were included, of which 10 reported on anxiety, 21 on depression and 23 on other indicators of mental ill-health. Overall, previous studies reported that mothers of people with IDs experienced poorer mental health as compared with mothers of typically developing people. Meta-analyses revealed significant findings for anxiety, depression, parenting stress, emotional burden and common mental disorders, but not for somatic symptoms. However, there was a considerable heterogeneity; hence, interpretation of results should be cautious. Identified gaps included scarce research on mental ill-health of mothers of adults with IDs at different stages of the caregiving trajectory.

Conclusions There is evidence of poorer mental ill-health in mothers of people with IDs compared with mothers of typically developing people, but lack of focus on different stages of the caregiving trajectory, methodological inconsistencies between studies and lack of robust studies pose limitations. This highlights the need both for improved support for mothers of people with IDs and for further methodologically robust research.

Keywords anxiety, depression, intellectual disabilities, mental health, mothers, systematic review
Background

Reviews of evidence on prevalence of mental ill-health

Mothers of children with intellectual disabilities (IDs) have been reported to have poorer mental health compared with other carer groups (e.g. those supporting a child with physical disabilities or an elderly parent with dementia) (Pinquart and Sörensen 2003). These mothers also appear to have more depressive, anxiety and stress symptoms than fathers of children with IDs (Singer 2006; Montes and Halterman 2007; Cairns et al. 2012, 2014; Giallo et al. 2015). However, most studies include small and highly selective samples, without comparison groups of parents of typically developing children (e.g. Chen et al. 2001; Azeem et al. 2013; Dawson et al. 2016; Rimmerman et al. 2018). We believe that research including large samples with comparison groups of mothers of typically developing people is important to undertake in view of health inequalities potentially associated with IDs. Should such associations be found, there would be implications for family systems and child development, particularly given the potential to then advocate for early preventative interventions.

A brief narrative overview of studies on mental health needs of parents with IDs reported a high prevalence of mental health needs in this population. However, the review was not systematic, and included studies without comparison groups of mothers of typically developing children (O’Keeffe and O’Hara 2008). A further systematic review found an association between positive impact of caring for a child with IDs and wellbeing in parents of children with IDs, with poor wellbeing accompanied by lower positive perceptions and higher negative perceptions of caregiving. However, this relationship was affected by extraneous variables including partner wellbeing, challenging behaviour, and syndrome characteristics (Horsley and Oliver 2015).

A research critique (Bailey et al. 2007) and two meta-analyses (Singer 2006; Miodrag et al. 2015) on maternal depression and stress in families of children with various disabilities or chronic health conditions reported that mothers of children with disabilities exhibited higher rates of depressive symptoms, stress and clinical levels of depression. A literature review of studies on stress, poor sleep and wellbeing in mothers of children with developmental disabilities found that they experienced more of these problems than mothers of typically developing children, and these problems were more persistent (Lee 2013). We did not locate any systematic reviews investigating prevalence of mental ill-health in mothers of people with IDs compared with mothers of typically developing children.

Mental ill-health at different stages of the caregiving trajectory

Improvements in health and social care and deinstitutionalisation mean that more people with IDs are living in the community and enjoying longer lives (Graham et al. 2013). Due to this prolonged caregiving role, parent carers of people with IDs face exceptional challenges and have very different circumstances from those people who come to care for relatives following onset of illness or trauma-related disability (Innes et al. 2012). Caregivers of children with IDs need more information on education and therapy (Jackson et al. 2016) in addition to help with parenting and coping (Douma et al. 2006; James 2013), especially if their child also experiences complex healthcare needs (Tan 2017). All of these factors might impact on mothers’ mental health in different ways at different stages of the caregiving trajectory (i.e. caring for a child, adult and older adult) (Emerson and Hatton 2011), but this is less clearly understood in the research. While caring can be an extremely positive and rewarding experience (Scorgie and Sobot 2000; Jokinen and Brown 2005), small-scale studies suggest it may also impact on parents’ mental health at key points in the caregiving trajectory (e.g. Chen et al. 2001; Cairns et al. 2014). Some research has directly compared mental health impacts on mothers at different ages, but has yet to compare impact on mothers with differently aged children. Chen et al. (2001) carried out a survey in the US looking at the health of 108 mothers of adults with IDs, divided into mid-life (younger) groups of women between 55 and 64 years of age and later-life (older) groups aged 65 years and over. The study found that caring for adult children with IDs might not be as detrimental to the mid-life mothers’ physical component of health as it might be to the later-life mothers. Both groups reported self-reported physical and mental health.
scores similar to the US national norms. Cairns et al. (2014) carried out a survey with 100 older parent carers subgrouped into young-old (65–74 years), middle-old (75–84 years) and old-old (85 + years) parent carers and found that mean mental health scores for the young-old (65–74) and middle-old (75–84) parent carers were below 50, that is, below the UK general population norms. While parent carers in the old-old (85+) group scored just above 50 (i.e. above average) compared with the two younger subgroups, the mean score for all three subgroups combined was still below UK norms. We did not locate any systematic reviews investigating prevalence of mental ill-health in mothers of people with IDs at different stages of the caregiving trajectory.

For these reasons, we aimed to systematically review mental ill-health in mothers of people with IDs compared with mothers of typically developing children and adults and investigate impact of caring on mental ill-health at different stages of the caregiving trajectory. Specific research questions were as follows:

1. How common is mental ill-health in mothers of people with IDs compared with mothers of typically developing people, and at different stages of the caregiving trajectory?
2. Are there gaps in the evidence base on mental ill-health in mothers of people with IDs compared with mothers of typically developing people, and at different stages of the caregiving trajectory?

**Methods**

This review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO, registration number: CRD42018088197). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist, flow diagram and guidelines were followed (Shamseer et al. 2015). Four databases were searched: Medline, Embase, CINAHL and PsycINFO in February 2020. Search terms included terms to capture IDs, combined with terms on mental health and parent carers (detailed list is included in Appendix 1). Studies were limited to English language. No time limits were applied.

**Study screening and selection**

Papers were initially screened on their title and abstract by the first reviewer (E. R.). A second reviewer (K. D.) read a random 10% of titles and abstracts to ensure the selection approach was systematic. Both reviewers were post-doctoral researchers. Any discrepancies were planned to be resolved through discussion, with further double reviewing planned if consistency was not reached by this stage. Relevant full texts were then reviewed for eligibility by the first reviewer (E. R.). For four papers for which there were doubts on inclusion, papers were read by two more reviewers (K. D. and D. K.) and were subsequently excluded from the data extraction stage. Data were then extracted from the selected papers by the first reviewer (E. R.), using a structured database, on author; publication year; country; population size and type; comparison group; definition of mental ill-health; measures used; findings; and study quality. Additionally, we reviewed the reference lists of all included studies to check if they referenced any relevant publications that were not identified through our electronic searches. Strict inclusion and exclusion criteria were used.

**Inclusion criteria**

- Mother caring for a child with IDs (IQ ≤ 70, or children described as having ‘intellectual disability’, ‘mental retardation’ or equivalent terms listed in our search terms);
- Mothers aged 16 years and above;
- Observational studies, for example, cohort, case–control and cross-sectional studies;
- Studies from peer-reviewed journals.

**Exclusion criteria**

- Data on mothers of children with IDs were not separately reported and were less than 50% of the sample;
- Studies without a comparison group of mothers of typically developing children;
- Syndrome specific studies (e.g. Down syndrome, Williams syndrome, Fragile X syndrome and autism);
- Studies on samples with developmental delay;
- Non-human studies;
- Grey literature.

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Quality assessment

The 12-item Critical Appraisal Skills Programme Checklist (CASP 2017) for cohort studies was used to systematically assess study quality. This covers all domains of quality assessment of observational studies (Sanderson et al. 2007), including clarity of the stated aims, methodology (including age/gender standardisation and consideration of group differences in disease prevalence rates), study design and size, participant selection, measures used, data collected, analyses employed, results, bias, generalisability, conflict of interests, and ethical procedures. Quality appraisal was double rated. First reviewer (E. R., research fellow) rated all papers while the second (K. D., research associate) and third (D. K., senior lecturer) reviewer second rated half of the reviewed papers each. Freedom from risk of bias was appraised per domain: ‘Yes’ (low risk), ‘Cannot tell’ (unknown risk) or ‘No’ (high risk). The following classification was used for risk of bias for each study overall (Mathie et al. 2017):

- Rating A = low risk of bias for all 12 items;
- Rating Bx = uncertain risk of bias for x items, low risk of bias in all other items;
- Rating Cx,y = high risk of bias in x items, uncertain risk of bias in y items and low risk of bias in all other items.

Meta-analysis of studies

Meta-analysis was undertaken using Review Manager (version 5.4). All outcome measures from the included studies were reported on a continuous scale. Only studies that reported on the mean, standard deviation and sample size for mothers of people with IDs and the comparison groups were included in the meta-analyses, as these statistics were necessary to calculate an unbiased standardised mean difference (SMD) between the studied populations. A negative mean difference indicated poorer mental health for the comparison group than for mothers of people with IDs. For papers that included more than one group of mothers of children with IDs (e.g. mothers of children with IDs and autism), but only one comparison group for typically developing population, we calculated a combined mean and standard deviation for these subgroups to minimise bias. Effect size was interpreted as small when SMD < 0.40, moderate when SMD ranged from 0.40 to 0.70 and large when SMD > 0.70 (Schünemann et al. 2011).

The Chi-squared statistic I² was chosen to measure the level of heterogeneity across the studies, as it allows for interpretation of results regardless of the number of studies included in the meta-analysis or their type of outcome data or effect measurement (Higgins et al. 2003). Heterogeneity was interpreted as not observed when I² = 0%, low when I² = 25%, medium when I² = 50% and high when I² = 75% (Higgins et al. 2003). Random effects models were selected for this analysis due to the different measures (e.g. different indicators of mental health) in the included studies. We intended to assess publication bias using funnel plot techniques and Egger’s regression test as appropriate, given the known limitations of these methods. The meta-analysis was performed on different subgroups, including anxiety, depression and other indicators of mental ill-health.

Sensitivity analysis

Sensitivity analysis was used to assess the impact of risk of bias for each study on the pooled SMD. Data were removed one-by-one from the meta-analysis for each study, beginning with the lowest ranked papers, to determine the effect of each individual study on the pooled SMD.

Results

The search returned 4613 records, of which 251 articles were read in full and 32 selected for inclusion and data extraction (Fig. 1). There were no disagreements between reviewers (κ = 1.00). Of the 32 studies, 16 had appropriate data for meta-analysis, although all meta-analyses revealed high heterogeneity, and hence, the meta-analysis results must be treated with considerable caution.

Table 1 presents main study characteristics. Four were from the UK, 10 from the USA, six from Sweden, two from Australia, two from Iran and eight from other countries. Twenty-six were from high-income countries (UK, USA, Australia, Sweden, Germany, Italy, Israel and Bahrain), four from upper middle-income countries (Iran, Malaysia and Turkey) and two in lower middle-income countries.
Publication dates were 1966–2019, with seven studies published before 2000. The majority included mothers whose children were under the age of 18 ($n = 29$); only two included mothers of both children and adults (both by the same author using the same Australian dataset), and one included mothers of adults only. Four out of 32 studies used unvalidated measures; a 46-item sentence completion test constructed by Cummings et al. (1966), self-rated health scale (Olsson and Hwang 2008), a 4-point scale on a self-completion questionnaire (Emerson et al. 2006) and a questionnaire from an unpublished preliminary study (Lenhard et al. 2005). A further two used ICD-9 and ICD-10 codes that had been recorded in health registries (Fairthorne et al. 2015; Fairthorne et al. 2016). Publication bias was not assessed as there were inadequate numbers of included studies to properly assess a funnel plot or conduct more advanced regression-based assessments.

We did not exclude papers based on quality, but quality appraisal revealed limitations to most of the studies. Small and selective samples, lack of precise reporting of findings or confounding factors in the design and analysis were the most common flaws (further details included in Appendix 2). Eight papers had low risk of bias, 12 had uncertain risk of bias and 12 presented high risk of bias (Table 1). There was a near perfect agreement between reviewers ($\kappa = 0.95$).

![Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart of the study selection process](image)
| Year  | Author            | Country | Mothers ID (N) | Mothers ID Age (years) | Mothers TD (N) | Mothers TD Age (years) | Mothers recruited from                                                                 | Risk of bias | Risk of bias rating |
|-------|-------------------|---------|----------------|------------------------|----------------|------------------------|----------------------------------------------------------------------------------------|--------------|---------------------|
| 1966  | Cummings et al.   | USA     | 60             | 60                     | 60             | 60                     | Clinics, social agencies and community physicians                                       | Low          | A                   |
| 1978  | Miller & Keirn    | USA     | 44             | M = 34.8               | 41             | M = 36.7               | University neuropsychiatric outpatient clinic and local newspaper/telephone recruitment | High         | C2.1                |
| 1989  | Harris & McHale   | USA     | 30             | M = 34.52, SD = 7.93   | 30             | M = 36.28, SD = 3.61   | Schools, social and medical services                                                   | Uncertain    | B2                  |
| 1989  | Walker et al.     | USA     | 24             | 24                     | 24             | 24                     | Special education schools and general paediatrics clinics records                      | Uncertain    | B1                  |
| 1992  | Walker et al.     | USA     | 24             | 24                     | 24             | 24                     | Special education schools and general paediatrics clinics records                      | High         | C2.1                |
| 1993  | Andersson         | Sweden  | 47             | M = 34                 | 47             | M = 34                 | Disability services and personal contacts                                              | High         | C2                  |
| 1999  | Ong et al.        | Malaysia| 75             | M = 36.3, SD = 6.1     | 75             | M = 37.1, SD = 6.6     | Paediatric hospital institute and outpatient clinic                                   | Uncertain    | B1                  |
| 2001  | Olsson & Hwang†   | Sweden  | 145            | 204                    | 204            | 204                    | Disability services and randomly selected control families living in the same area     | Low          | A                   |
| 2002  | Olsson & Hwang†   | Sweden  | 144            | 202                    | 202            | 202                    | Disability services and randomly selected control families living in the same area     | Uncertain    | B1                  |
| 2002  | Weiss             | USA     | 40             | 24–48 years old        | 40             | 24–48 years old        | Disability services, statewide developmental disabilities conference and parent networking organisation | Low         | A                   |
| 2003  | Emerson           | UK      | 245            | 9481                   | 9481           | 9481                   | Secondary data analysis of the ONS survey of the Mental Health of Children and Adolescents in Great Britain, 1999 | High         | C2.1                |
| 2005  | Eisenhower et al† | USA     | 43             | M = 36.0               | 136            | M = 35.0               | Disability services, preschools and day care centres                                  | Uncertain    | B1                  |
| 2005  | Lenhard et al†    | Germany | 66             | Age at child’s birth: M = 28.2, SD = 4.9 | 69             | Age at child’s birth: M = 31.5, SD = 4.3 | Special education and mainstream schools                                                | High         | C1.3                |
| 2006  | Abasibong et al.  | Nigeria | 106            | M = 40.0               | 101            | M = 34.8               | Disability services and higher education institution                                   | High         | C1.1                |
| Year | Author           | Country    | Mothers ID (N) | Mothers ID Age (years) | Mothers TD (N) | Mothers TD Age (years) | Mothers recruited from                                                                 | Risk of bias | Risk of bias rating |
|------|------------------|------------|----------------|------------------------|----------------|------------------------|---------------------------------------------------------------------------------------|-------------|---------------------|
| 2006 | Emerson et al.   | UK         | 514            | –                      | 6440           | –                      | Secondary analysis of the Department for Work and Pensions' Families & Children Study, Wave 4 | High         | C2.2                |
| 2006 | Olsson & Hwang   | Sweden     | 167            | M = 39.8               | 185            | M = 39.3               | Disability services and randomly selected control families living in the same area       | Uncertain    | B2                  |
| 2007 | Mugno et al.     | Italy      | 49             | –                      | 42             | –                      | Neurological and psychiatric centre, mainstream schools                                 | Low          | A                   |
| 2008 | Neece & Baker    | USA        | 74             | M = 36.1, SD = 6.5     | 115            | M = 37.9, SD = 5.9     | Community services for people with developmental disabilities, preschools and daycare programmes | Uncertain    | B1                  |
| 2010 | Olsson & Hwang   | Sweden     | 62             | –                      | 178            | –                      | Disability services and SPAR register of all individuals living in Sweden               | Uncertain    | B2                  |
| 2011 | Gupta & Kaur     | India      | 30             | –                      | 13             | –                      | Special education and mainstream schools                                               | High         | C3                  |
| 2011 | Mirsleih et al.  | Iran       | 124            | 25–50 years old        | 124            | 25–50 years old        | Special education and mainstream schools in southeastern Tehran                           | High         | C2                  |
| 2011 | Totsika et al.   | UK         | 412            | –                      | 14 444         | –                      | Secondary data analysis of the Millennium Cohort Study                                   | Uncertain    | B1                  |
| 2011 | Totsika et al.†  | UK         | 590            | >70% 30–44 years old   | 17 727         | >70% 30–44 years old   | Secondary analysis of two UK national surveys on psychiatric morbidity of 3–16-year-olds | Uncertain    | B2                  |
| 2013 | Kilic et al.     | Turkey     | Special education: 75; Mainstream education: 70 | 23–29 years N = 55; 30–36 years N = 48; 37 + years N = 42 | 75             | 23–29 years N = 29; 30–36 years N = 16; 37 + years N = 30 | Disability services                                                                     | High         | C1.1                |
| 2013 | Norlin & Broberg | Sweden     | T1: 58; T2: 46 | M = 34.0               | T1: 178; T2: 131| M = 33.8               | Disability services and SPAR register of all individuals living in Sweden               | Low          | A                   |
| 2015 | Long et al.      | USA        | Latina: 48; non-Latina: 50 | Latina: M = 36.58, SD = 5.61; non-Latina: M = 42.02, SD = 5.18 | Latina: 49; non-Latina: 45 | Latina: M = 38.68, SD = 6.00; non-Latina: M = 39.78, SD = 6.52 | Hospital-based general and specialty paediatric programmes, community agencies, paediatricians' offices, public and private schools, and word of mouth | Uncertain    | B1                  |
| 2018 | AlAnsari and Jahrami | Bahrain     | 30             | M = 33.3, SD = 6.5     | 30             | M = 33.3, SD = 6.5     | National intellectual disability/autism registry and social contacts of mothers in ID group | High         | C2.1                |
Table 1. (Continued)

| Year | Author | Country | Mothers ID (N) | Mothers ID Age (years) | Mothers TD (N) | Mothers TD Age (years) | Mothers recruited from | Risk of bias | Risk of bias rating |
|------|--------|---------|----------------|------------------------|----------------|------------------------|-----------------------|-------------|---------------------|
| 2018 | Hosseinik et al. | Iran | Conflicting sample sizes (94, 40 and 48) reported in the abstract, methods and tables | -- | Conflicting sample sizes (94, 40 and 48) reported in the abstract, methods and tables | -- | Special education schools and mothers of TD children residing in Yasouj city | High | C5.2 |
| 2019 | Blacher & Baker | USA | 28 | M = 43.3 | 84 | M = 45.5 | Diagnostic/intervention services for people with developmental disabilities, preschools, schools and daycare programmes | Low | A |

Studies of mothers of children and adults

| Year | Author | Country | Mild–moderate ID: 4629; Severe ID: 322 | Age at index birth: mild–moderate ID: | Age at index birth: severe ID: | Population of women who gave birth to a live child in Western Australia between Jan 1983 and Dec 2005 | Risk of bias | Risk of bias rating |
|------|--------|---------|-----------------|-----------------|-----------------|-----------------------------|-------------|---------------------|
| 2015 | Fairthorne et al. | Australia | 271 249 | <20 years N = 519; 20–34 years N = 3668; >35 years N = 492; severe ID: <20 years N = 24; 20–34 years N = 249; >35 years N = 49 | <20 years N = 19 764; 20–34 years N = 22 1229; >35 years N = 32 256 | | Low | A |

| Year | Author | Country | Mild–moderate ID: 4629; Severe ID: 322 | Age at index birth: mild–moderate ID: | Age at index birth: severe ID: | Population of women who gave birth to a live child in Western Australia between Jan 1983 and Dec 2005 | Risk of bias | Risk of bias rating |
|------|--------|---------|-----------------|-----------------|-----------------|-----------------------------|-------------|---------------------|
| 2016 | Fairthorne et al. | Australia | 271 249 | <20 years N = 637; 20–34 years N = 4687; >35 years N = 603 | <20 years N = 22 085; 20–34 years N = 235 820; >35 years N = 32 679 | | Low | A |

Studies of mothers of adults

| Year | Author | Country | Population of women who gave birth to a live child in Western Australia between Jan 1983 and Dec 2005 | Risk of bias | Risk of bias rating |
|------|--------|---------|-----------------------------|-------------|---------------------|
| 2001 | Rimmerman & Muraver | Israel | Sheltered employment programme | Uncertain | B1 |

Abbreviations: DS, Down syndrome; ID, children with intellectual disability; M, mean; SD, standard deviation; TD, typically developing children.

These papers additionally separately analysed subgroups of people with intellectual disabilities and Down syndrome and/or intellectual disabilities and autism.
Table 2: Studies investigating anxiety in mothers of children with intellectual disabilities

| Year   | Author          | Mothers ID | Mothers TD | Statistical comparison | Measures used         | Definition of mental ill-health |
|--------|-----------------|------------|------------|------------------------|-----------------------|--------------------------------|
| 1978   | Miller & Keirn  | M = 52.63, SD = 8.1 | M = 52.35, SD = 7.24 | Not reported            | MMPI                  | Elevations reflect parents’ pathological reactions to the presence of a disabled child; psychasthenia scale |
| 1993   | Andersson       | Short experience: M = 7.03; long experience: M = 6.72; total: M = 6.92 | Short experience: M = 7.14; long experience: M = 6.71; total: M = 6.98 | No statistically significant differences found between the groups | HADS                  | HADS: anxiety score ≥ 8 |
| 2002   | Weiss           | M = 14.4, SD = 2.82 | M = 13.2, SD = 2.65 | F = 3.45, P = .035     | Questionnaire developed by the US Department of Health, Education and Welfare | 7 questions on anxiety-related (e.g. jitteriness, nervousness, irritability, anger) symptoms |
| 2003   | Emerson         | 36.0% worried to a great extent; 34.0% to some extent | 5.0% worried to a great extent; 15.0% to some extent | Mann–Whitney z = 19.61, P < .001 | SDQ                  | Self-assessed psychological impact of the difficulties of the sampled child/adolescent on the respondent |
| 2005   | Lenhard et al.  | 51.9% had a significantly elevated score | 31.8% had a significantly elevated score in ID mothers compared with TD mothers (P = .004) | STAI                  | STAI: score > 60 |
| 2006   | Abasiubong et al. | N = 27 (25.5%) with score ≥ 8 | N = 7 (6.9%) with score ≥ 8 | t = 4.79, P < .001 | GHQ-28 | GHQ-28: symptoms of anxiety/insomnia; higher score indicates worse health |
| 2011   | Mirsaleh et al. | M = 7.88, SD = 4.62 | M = 5.56, SD = 2.79 | F = 8.579, P < .001     | STAI                  | STAI: score > 60 |
| 2013   | Kilic et al.    | X = 49.87, SS = 6.5 | X = 49.87, SS = 6.5 | t = 4.79, P < .001     | STAI                  | STAI: score > 60 |
| 2018   | AlAnsari and Jahrami | M = 2.43, SD = 0.4 | M = 1.03, SD = 0.27 | P = .008 | GHQ-28 | GHQ-28: symptoms of anxiety/insomnia; higher score indicates worse health |
| 2015   | Fairthorne et al. | Neurotic disorders: mild–moderate ID: 326 (7.0%); severe ID: 19 (5.9%) | Neurotic disorders: 8038 (3.0%) | Mild–moderate ID: IRR = 1.80, 95% CI [1.3, 2.5]; severe ID: IRR = 1.98, 95% CI [0.6, 6.4] | ICD-9 and ICD-10 codes | Statutory state-based registries of hospital admissions and outpatient mental health appointments |

Abbreviations: CES-D, Centre for Epidemiological Studies Depression Scale; DIGS, Diagnostic Interview for Genetic Studies; GHQ, General Health Questionnaire; HADS, Hospital Anxiety and Depression Scale; ID, children with intellectual disability; M, mean score; MMPI, Minnesota Multiphasic Personality Inventory; SD, standard deviation; SS, sum of squared deviations; STAI, State–Trait Anxiety Inventory; TD, typically developing children.
Anxiety

Ten studies investigated anxiety (Table 2); nine on mothers of children and one on mothers of children and adults. Two studies used each of the following measures: Hospital Anxiety and Depression Scale (Andersson 1993; Abasiubong et al. 2006), State–Trait Anxiety Inventory (Lenhard et al. 2005; Kilic et al. 2013) and General Health Questionnaire, which has an anxiety sub-scale (Mirsaleh et al. 2011; AlAnsari and Jahrami 2018). The remaining four studies used a variety of other instruments (Miller and Keirn 1978; Weiss 2002; Emerson 2003; Fairthorne et al. 2015). All studies apart from two (Miller and Keirn 1978; Andersson 1993) reported more anxiety in mothers of children with IDs. The biggest differences were observed by Abasiubong et al. (2006), Emerson (2003), and Lenhard et al. (2005). Rates of anxiety in the secondary analysis of routinely collected administrative data on mothers of both children and adults (Fairthorne et al. 2015) were lower than in some studies of mothers of children only (e.g. Lenhard et al. 2005), but direct comparisons cannot be made due to differing methodological approaches.

Data were suitable for meta-analysis in four studies (Miller and Keirn 1978; Weiss 2002; Mirsaleh et al. 2011; AlAnsari and Jahrami 2018) (Fig. 2). The pooled SMD for differences in anxiety levels between mothers of children with IDs and mothers of typically developing children was statistically significant at \( t = 1.18, 95\% \text{ CI} [0.18, 2.17], P < .05 \). There was a large effect size, demonstrating that mothers of children with IDs experience heightened anxiety compared with mothers of typically developing children. Heterogeneity between studies was, however, at a high level, with \( I^2 = 95\% \), and the significant finding was largely driven by the study by AlAnsari and Jahrami (2018).

| Study or Subgroup               | Mothers ID Mean | SD    | Total  | Mothers TD Mean | SD    | Total  | Weight |
|--------------------------------|----------------|-------|--------|----------------|-------|--------|--------|
| AlAnsari & Jahrami 2018        | 2.43           | 0.4   | 30     | 1.03           | 0.27  | 30     | 22.1%  |
| Miller & Keirn 1978            | 52.63          | 8.11  | 44     | 52.35          | 7.24  | 41     | 25.7%  |
| Mirsaleh et al. 2011           | 7.88           | 4.62  | 124    | 5.56           | 2.79  | 124    | 26.5%  |
| Weiss 2002                     | 14.4           | 2.82  | 40     | 13.2           | 2.65  | 40     | 25.6%  |
| Total (95\% CI)                | 238            | 235   | 100.0% | 1.18 [0.18, 2.17] |

Heterogeneity: \( \chi^2 = 0.95; \text{ df} = 3 (P < 0.00001); I^2 = 95\% \)

Test for overall effect: \( Z = 2.32 (P = 0.02) \)

Figure 2. Forest plot for four analysable studies presenting findings on anxiety in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

Depression

Twenty-one studies investigated depression (Table 3); 20 studies on mothers of children and one on mothers of children and adults. Eight used the Beck Depression Inventory (Harris and McHale 1989; Kilic et al. 2011; Norlin and Broberg 2013; Olsson and Hwang 2001, 2002, 2006, 2008; Weiss 2002), two used the Centre for Epidemiological Studies Depression Scale (Walker et al. 1989; Eisenhower et al. 2005), Hospital Anxiety and Depression Scale (Andersson 1993; Abasiubong et al. 2006) and General Health Questionnaire, which has a depression sub-scale (Mirsaleh et al. 2011; AlAnsari and Jahrami 2018). Seven remaining papers used a variety of other instruments (Cummings et al. 1966; Miller and Keirn 1978; Ong et al. 1999; Emerson 2003; Emerson et al. 2006; Fairthorne et al. 2015; Long et al. 2015).

All studies reported higher depression scores in mothers of children with IDs, except one reporting statistically non-significant lower scores in mothers of children with IDs (Walker et al. 1989). When Emerson et al. (2006) controlled for maternal characteristics (i.e. age, marital status, general health status and health problems limiting activity), the association between caring for a child with IDs and maternal happiness was no longer statistically significant.

Data were suitable for meta-analysis in 13 studies (AlAnsari and Jahrami 2018; Cummings et al. 1966; Eisenhower et al. 2005; Harris and McHale 1989; Miller and Keirn 1978; Mirsaleh et al. 2011; Norlin and Broberg 2013; Olsson and Hwang 2002, 2006, 2008; Ong et al. 1999; Walker et al. 1989; Weiss 2002) (Fig. 3). Norlin and Broberg (2013) collected data at two time points. Due to a decrease in number of participants at time 2, we meta-analysed data for time...
Table 3: Studies investigating depression in mothers of children with intellectual disabilities

| Year | Author               | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|----------------------|------------|------------|------------------------|---------------|---------------------------------|
| 1966 | Cummings et al.      | M = 7.3, SD = 2.0 | M = 5.3, SD = 2.1 | t = 5.26, P = .01 | Bespoke Sentence Completion Test | Level of depressive feeling; higher score indicates higher level |
| 1978 | Miller & Keirn       | M = 56.23, SD = 10.33 | M = 53.24, SD = 7.30 | Not reported | MMPI | Elevations reflect parents' pathological reactions to the presence of a disabled child; depression scale |
| 1989 | Harris & McHale      | M = 5.77, SD = 5.00 | M = 3.87, SD = 4.24 | No significant group differences, but ID mothers reported somewhat higher depression; the level of symptomology ranged from no depression to mild depression | BDI | Score range of 0–63, with higher scores representing more intense depressive symptomology |
| 1989 | Walker et al.        | M = 9.67, SD = 9.75 | M = 10.12, SD = 10.08 | F3,91 = 0.39, P not statistically significant; ID: β = −0.03 (8 = −.73), P not reported | CES-D | CES-D questionnaire to measure depressive symptoms in adults, 20 items, 4-point scale, score range 0–60, score > 16 for depression |
| 1993 | Andersson            | M = 4.30 | M = 3.60 | No statistically significant differences found between the groups P < .001 | HADS | HADS: depression score ≥8 |
| 1999 | Ong et al.           | M = 24.7, SD = 5.8 | M = 21.5, SD = 3.5 | Not reported | BDI | Child domain (CDS) and parent-domain stress (PDS), Life Stress and a total PSI score BDI: 4-point scale of 0–4 with score 10–20 for dysphoria and >20 for depression |
| 2001 | Olsson & Hwang       | M = 9.2, dysphoria N = 53 (37.0%), depression N = 12 (8.0%), dysphoria N = 7 (4.0%) | M = 5.2, dysphoria N = 27 (13.0%), depression N = 7 (4.0%) | Correlation between SoC and BDI: ID: r = −0.72, P < .01 | SoC | SoC: 7-point scale, 13 items, e.g. feeling of not being in control, of no meaning to things, of disappointment, higher score indicates higher sense of coherence |
| 2002 | Olsson & Hwang       | 1. M = 9.2, SD = 7.4 | 1. M = 5.2, SD = 5.2 | 2. Low SoC M = 9.7 | 1. BDI | BDI: 4-point scale of 0–4 with score 10–20 for dysphoria and >20 for depression |
|      | 2. Low SoC M = 4.5 | 2. High SoC M = 23 (26.0%), Depression low SoC N = 7 (8.0%), Depression high SoC N = 0 (0%) | 2. High SoC M = 23 (26.0%), Depression low SoC N = 7 (8.0%), Depression high SoC N = 0 (0%) | When adjusted for SoC, fewer ID mothers with low SoC had normal BDI scores compared with TD mothers: F2,216 = 9.0, P < .01 |
| 2002 | Weiss                | M = 15.5, SD = 2.81 | M = 9.2, SD = 2.16 | F = 15.95, P < .001 | BDI | BDI: symptoms of depression (e.g. hopelessness, loss of interest in activities, changes in diet and sleep) |

BDI: symptoms of depression
SoC: 7-point scale, 13 items, e.g. feeling of not being in control, of no meaning to things, of disappointment, higher score indicates higher sense of coherence
| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|---------------|---------------------------------|
| 2003 | Emerson | Depressed to a great extent 14.0%; to some extent 30.0% | Depressed to a great extent 2.0%; to some extent 7.0% | Mann–Whitney $z = 17.93$, $P < .001$ | SDQ | Self-assessed psychological impact of the difficulties of the sampled child/adolescent on the respondent |
| 2005 | Eisenhower et al. | $N = 11$ (10.4%) with score $\geq 8$ | $N = 4$ (3.9%) with score $\geq 8$ | Not reported | CES-D | CES-D questionnaire to measure depressive symptoms in adults; higher scores indicate higher level of depressive symptoms |
| 2006 | Abasibong et al. | Depression in relation to participation in work: <20 h/week $M = 11.4$, $SD = 10.0$; 20–33 h/week $M = 8.8$, $SD = 6.5$; >33 h/week $M = 8.2$, $SD = 6.7$ | Depression in relation to participation in work: <20 h/week $M = 7.8$, $SD = 7.5$; 20–33 h/week $M = 5.0$, $SD = 5.8$; >33 h/week $M = 4.3$, $SD = 5.2$ | Not reported | HADS | HADS: depression score $\geq 8$ |
| 2006 | Emerson et al. | Happy: not at all 1.2%; not very 83% | Happy: not at all 0.5%; not very 3.3% | Mothers of children with ID significantly less happy: Mann–Whitney $z = 4.71$, $P < .001$. Association between caring for a child with ID and maternal happiness: OR = 1.42, $P < .01$. Impact of controlling for maternal characteristics on the association between caring for a child with ID and maternal happiness: OR = 1.00, $P$ not reported. Maternal general health not good: OR = 2.24, $P < .001$ | 4-point scale on a self-completion questionnaire | Self-report of feeling unhappy |
| 2008 | Olsson & Hwang | $I. M = -12.4$, $SD = 13.9$ | $I. M = -20.9$, $SD = 12.9$ | $I. More ID mothers scored above the cut-off point for low wellbeing $\lambda (2) = 16.5$, $P = .01$ | $I. BDI-2r$, 7-point scale of −3 (could not be happier) to 3 (could not be more unhappy) | High wellbeing (BDI $–61$–9) Medium wellbeing (BDI $–8$–8) Low wellbeing (BDI $9$–61) |
| Year     | Author                      | Mothers ID                | Mothers TD                | Statistical comparison                                                                 | Measures used               | Definition of mental ill-health                                                                 |
|----------|-----------------------------|---------------------------|---------------------------|----------------------------------------------------------------------------------------|-----------------------------|-----------------------------------------------------------------------------------------------|
| 2011     | Mirsaleh et al.             | M = 4.52, SD = 4.98       | M = 1.43, SD = 1.98       | t = 6.42, P < .001                                                                      | GHQ-28                      | GHQ-28: symptoms of severe depression; higher score indicates worse health                      |
| 2013     | Kilic et al.                | X = 14.77, SS = 6.9       | F = 3.521, P = .031       | BDI                                                                                     | BDI: not reported           |                                                                                               |
| 2013     | Norlin & Broberg            | T1: M = –12.4, SD = 13.8  | T1: M = –20.8, SD = 12.9  | ID vs. TD mothers: T1: F = 10.18, P < .001 T2: not reported for mothers separately. Child ID status contributed significantly to wellbeing in mothers t = 3.18, P < .01 | BDI-2r                      | BDI-2r: 7-point scale of –3 (could not be happier) to 3 (could not be more unhappy); range –63 to 63 with higher scores for poorer wellbeing |
| 2015     | Long et al.                 | Not reported              | Not reported              | Significant group differences with highest levels of depressive symptoms observed in Latina ID mothers: F<sub>3,188</sub> = 4.92, P = .003 | BSI: depressive symptoms scale | 5-point scale                                                                                  |
| 2018     | AlAnsari and Jahrami        | M = 1.46, SD = 0.3        | M = 0.86, SD = 0.19       | P = .270                                                                                | GHQ-28                      | GHQ-28: symptoms of severe depression; higher score indicates worse health                      |
| 2015     | Fairthorne et al.           | Affective disorders: mild–moderate ID: 7881 (2.9%) | Affective disorders: mild–moderate ID: 7881 (2.9%) | ICD-9 and ICD-10 codes                                                                 | Statutory state-based registries of hospital admissions and outpatient mental health appointments |
|          | Studies of mothers of children and adults | | | | | |

Abbreviations: BDI, Beck Depression Inventory; BSI, Brief Symptom Inventory; CES-D, Centre for Epidemiological Studies Depression Scale; DIGS, Diagnostic Interview for Genetic Studies; GHQ, General Health Questionnaire; HADS, Hospital Anxiety and Depression Scale; ID, Children with intellectual disability; M, mean score; MMPI, Minnesota Multiphasic Personality Inventory; PSI, Parenting Stress Index; SD, standard deviation; SDQ, Strengths and Difficulties Questionnaire; SMI, serious mental illness; SoC, Sense of Coherence Scale; SS, sum of squared deviations; TD, typically developing children.
For two papers that included more than one group of mothers of children with IDs (Olsson and Hwang 2002; Eisenhower et al. 2005), but only one comparison group for the typically developing population, we calculated a combined mean and standard deviation for these subgroups to minimise bias. For Olsson and Hwang (2006), only the results for the total control and comparison group were entered in the meta-analysis. The remaining results were presented by three categories of work hours and were not directly related to the study’s hypothesis testing if a presence of a child with ID in the family is related to parental wellbeing. The pooled SMD for differences in depression scores between mothers of children with IDs and mothers of typically developing children was 0.77, 95% CI [0.52, 1.02], $P < .001$, showing that mothers of children with IDs have higher depression than mothers of typically developing children. However, there was a high level of heterogeneity between studies, with $I^2 = 86\%$. Due to substantial heterogeneity, a further meta-analysis was then conducted with the six eligible papers that had used the Beck Depression Inventory (Harris and McHale 1989; Norlin and Broberg 2013; Olsson and Hwang 2002, 2006, 2008; Weiss 2002), with similar results: effect size was 0.84 95% CI [0.50, 1.18], $P < .001$, with high heterogeneity: $I^2 = 87\%$.

Other indicators of mental ill-health

Twenty-three studies investigated other indicators of mental ill-health (Table 4); 20 on mothers of children, two on mothers of children and adults, and one on mothers of adults. Five used the General Health Questionnaire (Emerson 2003; Abasiubong et al. 2006; Mirsaleh et al. 2011; Totsika et al. 2011b; AlAnsari and Jahrami 2018), four used the Family Impact Questionnaire (Eisenhower et al. 2005; Neece and Baker 2008; Norlin and Broberg 2013; Blacher and Baker 2019) and two used records in statutory state-based registries of hospital admissions and outpatient mental health appointments (Fairthorne et al. 2015, 2016). Fifteen studies used a variety of other instruments, sometimes in conjunction with those already mentioned. The studies have been divided into the categories of stress, emotional burden, common mental disorders, somatic symptoms, psychiatric disorders and wellbeing.

Stress

Stress was investigated in eight studies on mothers of children (Ong et al. 1999; Eisenhower et al. 2005; Neece and Baker 2008; Gupta and Kaur 2010; Totsika et al. 2011a; Norlin and Broberg 2013; Hosseininik et al. 2018; Blacher and Baker 2019). Four used the Family Impact Questionnaire (Eisenhower et al. 2005; Neece and Baker 2008; Norlin and Broberg 2013; Blacher and Baker 2019). The remaining studies used a variety of measures.

Data were suitable for meta-analysis in six studies, which measured stress (Ong et al. 1999; Eisenhower et al. 2005; Gupta and Kaur 2010; Norlin and Broberg 2013; Hosseininik et al. 2018; Blacher and Baker 2019).

Figure 3. Forest plot for 13 analysable studies presenting findings on depression in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]
| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|--------------|--------------------------------|
| 1999 | Ong et al. | Total PSI: $M = 274$; SD = 42.5 | Total PSI: $M = 232.1$, SD = 31.8 | Total PSI: $P < .001$ | PSI | Child domain (CDS) and parent-domain stress (PDS), life stress and a total PSI score |
|      |        | Child domain: $M = 132.9$, SD = 23.6 | Child domain: $M = 106.8$, SD = 15.7 | Child domain: $P < .001$ |        | |
|      |        | Parent domain: $M = 141.4$, SD = 26.1 | Parent domain: $M = 126.5$, SD = 16.8 | Parent domain: $P < .001$ |        | |
|      |        | Social isolation: $M = 14.1$, SD = 3.9 | Social isolation: $M = 13.5$, SD = 2.4 | Social isolation: $P = .271$ |        | |
|      |        | Health problems: $M = 12.4$, SD = 3.3 | Health problems: $M = 11.5$, SD = 2.4 | Health problems: $P = .058$ |        | |
|      |        | Life stress: $M = 8.7$, SD = 7.9 | Life stress: $M = 7.5$, SD = 2.4 | Life stress: $P = .338$ |        | |
| 2005 | Eisenhower et al. | FIQ negative impact | FIQ negative impact | $B = -1.47$, SE = 2.52, $\beta = -0.06$ | FIQ | FIQ social relationships and negative feelings about parenting subscale combined score with higher scores indicating higher level of stress |
|      |        | $M = 18.3$, SD = 11.5 | $M = 11.1$, SD = 8.0 | |
| 2008 | Neece & Baker | Not reported | Not reported | Negative impact subscale: ID mothers reported more parenting stress at child's age 6 ($t = 4.41$, $P < .001$) and at child's age 8 ($t = 7.06$, $P < .001$) | FIQ | |
|      |        | FIQ | | |
| 2010 | Gupta & Kaur | Mental stress > physical stress in 21/30 mothers; highest mental stress score for all parents was 32; total stress score for all ID parents: $M = 22.16$, SD = 10.80 | Mental stress > physical stress in 7/13 mothers; highest mental stress score for all parents was 26; total stress score for all ID parents: $M = 20.50$, SD = 5.00 | Total stress score: $t = 2.46$; $P < .01$ | QSAT | Maximum possible score for mental health is 38; stressor occurs more than once a week = 2, every month = 1, less frequently than once a month = 0 |
|      |        | 1. SMI: 3.18% | 1. SMI: ID vs. TD $F = 32.57$, $P < .001$ | 1. K6 |        | |
|      |        | 2. Maternal psych. distress: 8.09% | 2. Maternal psych. distress ID vs. TD $F = 10.47$, $P < .001$ | 2. SF-8 Mental Component Summary |        | |
| 2011a | Totsika et al. | 1. K6 | 1. K6: self-report of psychological distress on a scale 0–4 with cut-off of ≥13 for SMI |        |        | |
|      |        | 2. SF-8 Mental Component Summary | 2. SF-8: mental health functioning on 1–6 scale, lower scores indicate frequent psychological distress and higher scores indicate frequent positive affect |        |        | |
| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|---------------|--------------------------------|
| 2013 | Norlin & Broberg | Parenting stress: $M = 10.8$, SD = 8.2 | Parenting stress: $M = 4.6$, SD = 3.8 | Group differences not reported for mothers separately | FIQ (16/50 items) | FIQ: 4-point scale from not at all to very much; higher score indicates higher level of stress |
| 2018 | Hosseininik et al. | Tolerance $M = 7$, SD = 1.16 | Tolerance $M = 7.50$, SD = 0.96 | Difference in at least one of the components: $F_{1,91} = 41.91, P = .001$ | DTS | Emotional distress tolerance self-evaluation index with 4 subscales |
| 2018 | | Absorption $M = 5$, SD = 1.16 | Absorption $M = 5.33$, SD = 0.95 | Tolerance: $F_{1,94} = 5.22, P = .025$ | | |
| 2018 | | Evaluation $M = 9.33$, SD = 0.95 | Evaluation $M = 9.83$, SD = 0.90 | Absorption: $F_{1,94} = 2.35, P = .12$ | | |
| 2018 | | Adaptation $M = 9.50$, SD = 0.96 | Adaptation $M = 10$, SD = 0.82 | Evaluation: $F_{1,94} = 6.94, P = .01$ | | |
| 2019 | Blacher & Baker | $M = 2.16, SD = 14.2$ | $M = 10.0, SD = 8.4$ | $F = 29.94, P < .001$ | FIQ | FIQ: 4-point scale from not at all to very much; higher score indicates higher level of stress |

Emotional burden
Studies of mothers of children
1992 Walker et al. | Personal burden: $M = 3.62$, SD = 1.28 | Personal burden: $M = 2.83$, SD = 1.27; remaining scales not administered to mothers of TD children because they refer to care of a family member with a chronic condition | Significant group differences found for: life-span care $F_{2,43} = 21.94, P < .001$; terminal illness stress $F_{2,43} = 30.99, P < .001$; total score $F_{2,43} = 4.72, P < .01$. Correlation of ID with: personal burden $r = .15, P$ not reported; life-span care $r = .35, P < .001$; lack of personal reward $r = -.12, P$ not reported; terminal illness stress $r = -.22, P < .05$; preference for institutional care $r = -.25, P < .05$. Correlations of depression with: personal burden $r = .50, P < .001$; life-span care $r = .16, P$ not reported; lack of personal reward $r = -.07, P$ not reported; terminal illness stress $r = .30, P < .01$; preference for institutional care $r = -.20, P < .05$; ID $r = -.16, P$ not reported | QRS-S | QRS-S 5 scales on respondent attitudes with 0–6 score range per scale |
| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|---------------|--------------------------------|
| 2002 | Weiss  | Emotional exhaustion: $M = 27.75, SD = 2.42$ | Emotional exhaustion: $M = 23.75, SD = 2.95$ | Emotional exhaustion: $F = 3.83, P = .024$ Mann-Whitney $z = 20.17, P < .001$ | MBI | Emotion emotional exhaustion subscale |
| 2003 | Emerson| Tiredness to a great extent 24.0%; to some extent 31.0% | Not reported | | SDQ | Self-assessed psychological impact of the difficulties of the sampled child/adolescent on the respondent |
| 2005 | Lenhard et al. | Not reported | Not reported | Emotional strain: significantly elevated score in ID mothers compared with the TD mothers ($P = .010$) | Questionnaire from an unpublished preliminary study | 6-point-scale ranging from 1 (totally disagree) to 6 (totally agree); subscale scores were computed with values ranging between 0 and 1 |

**Common mental disorders**

**Studies of mothers of children**

| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|---------------|--------------------------------|
| 2003 | Emerson | $M = 2.6$; 35% with GHQ $> 2$ | $M = 1.8$; 25% with GHQ score $> 2$ | Mann–Whitney $z = 13.9$, $df = 1; P < .001$ | GHQ-12 | GHQ $> 2$ |
| 2006 | Abasiubong et al. | $N = 28$ (26.4%) with GHQ score $> 5$ | $N = 10$ (9.9%) with GHQ score $> 5$ | Not reported | GHQ-28 | GHQ-28: score $> 5$ |
| 2007 | Mugno et al. | Psychological subdomain: $M = 68.79, SD = 13.40$ | Psychological subdomain: $M = 64.38, SD = 15.40$ | Psychological subdomain: $F = 4.17, P = .007$ | WHOQOL-BREF | Psychological subdomain: self-perception of mental state and wellbeing; higher score indicates better quality of life in psychological domain |
| 2011 | Mirsaleh et al. | Total score: $M = 27.29, SD = 14.73$ | Total score: $M = 17.49, SD = 7.07$ | Total score: $t = 6.68, P < .001$ | GHQ-28 | GHQ-28: total score; higher score indicates worse health |
| 2011b | Totsika et al. | Maternal emotional disorder: 32.4% | Maternal emotional disorder: 23.7% | 1D presence compared with TD OR = 1.02, 95% CI [0.84–1.24]; $\chi^2 = 40.12, P < .001$ $V = 0.047$ | GHQ-12 | GHQ-12: screening for common mental disorders |
| 2018 | AlAnsari and Jahromi | Total score: $M = 6.8, SD = 1.1$ | Total score: $M = 4.6, SD = 0.90$ | Not reported | GHQ-28 | GHQ-28: total score; higher score indicates worse health |
| 2019 | Blacher & Baker | $M = 25.9, SD = 24.5$ | $M = 14.2, SD = 14.3$ | $F = 13.19, P < .001$ | SCL | SCL: 4-point scale from not at all to extremely; higher score indicates higher level of adult mental health symptoms (somatisation, interpersonal sensitivity, anxiety, depression, and hostility); total score range is 0–140 |

[Table 4](#) (Continued)
| Year | Author | Mothers ID | Mothers TD | Statistical comparison | Measures used | Definition of mental ill-health |
|------|--------|------------|------------|------------------------|---------------|--------------------------------|
| 2002 | Weiss  | Somatic complaints: $M = 15.09, SD = 3.15$ | Somatic complaints: $M = 13.41, SD = 2.55$ | Somatic complaints: $F = 13.06, P < .001$ | Questionnaire developed by the US Department of Health, Education and Welfare | 10 questions on stress-related symptoms (e.g. sweaty hands, tachycardia and shortness of breath) |
| 2011 | Mirsaleh et al. | Somatic symptoms: $M = 7.06, SD = 4.08$ | Somatic symptoms: $M = 4.40, SD = 2.95$ | Somatic symptoms: $t = 5.89, P < .001$ | GHQ-28 | GHQ-28: somatic symptoms subscale; higher score indicates worse health |
| 2015 | Long et al. | Not reported | Not reported | Significant group differences with highest levels of GSI and somatic symptoms observed in Latina ID mothers: GSI $F_{1,188} = 3.30, P = .02$; somatic symptoms $F_{3,188} = 3.98, P = .009$ | BSI: Global Severity Index (GSI) and somatic symptoms scales | 5-point scale |
| 2018 | AlAnsari and Jahrami | Somatic symptom: $M = 1.13, SD = 0.3$ | Somatic symptom: $M = 1.46, SD = 0.39$ | Somatic symptom: $P = .501$ | GHQ-28 | GHQ-28: somatic symptoms subscale; higher score indicates worse health |
| 1978 | Miller & Keirn | Hypochondriasis: $M = 52.75, SD = 10.37$ | Hypochondriasis: $M = 53.40, SD = 9.73$ | Hypochondriasis: $t = 2.93, P < .005$; t-test results not reported for other scales | MMPI | Elevations reflect parents' pathological reactions to the presence of a disabled child; higher score indicates worse mental health |

### Somatic symptoms

- Studies of mothers of children

#### 2002
- Weiss
  - Somatic complaints: $M = 15.09, SD = 3.15$
  - Somatic complaints: $M = 13.41, SD = 2.55$
  - Somatic complaints: $F = 13.06, P < .001$

#### 2011
- Mirsaleh et al.
  - Somatic symptoms: $M = 7.06, SD = 4.08$
  - Somatic symptoms: $M = 4.40, SD = 2.95$
  - Somatic symptoms: $t = 5.89, P < .001$

#### 2015
- Long et al.
  - Not reported

#### 2018
- AlAnsari and Jahrami
  - Somatic symptom: $M = 1.13, SD = 0.3$

### Psychiatric disorders

#### 1978
- Miller & Keirn
  - Hypochondriasis: $M = 52.75, SD = 10.37$
  - Hypochondriasis: $M = 53.40, SD = 9.73$
  - Hypochondriasis: $t = 2.93, P < .005$; t-test results not reported for other scales

#### 1978
- Miller & Keirn
  - Hysteria: $M = 59.25, SD = 10.95$
  - Hysteria: $M = 58.19, SD = 7.93$
  - $t < 2.93, P < .005$; t-test results not reported for other scales

#### 1978
- Miller & Keirn
  - Psychopathic deviate: $M = 58.91, SD = 11.16$
  - Psychopathic deviate: $M = 56.84, SD = 7.15$
  - $t < 2.93, P < .005$; t-test results not reported for other scales

#### 1978
- Miller & Keirn
  - Paranoia: $M = 54.90, SD = 8.97$
  - Paranoia: $M = 55.05, SD = 8.54$
  - $t < 2.93, P < .005$; t-test results not reported for other scales

#### 1978
- Miller & Keirn
  - Schizophrenia: $M = 55.73, SD = 8.89$
  - Schizophrenia: $M = 34.76, SD = 7.05$
  - Schizophrenia: $M = 50.08, SD = 7.94$
  - $t < 2.93, P < .005$; t-test results not reported for other scales
| Year   | Author            | Mothers ID                                                                 | Mothers TD                                                                 | Statistical comparison                                                                 | Measures used         | Definition of mental ill-health                                                                 |
|--------|-------------------|----------------------------------------------------------------------------|----------------------------------------------------------------------------|----------------------------------------------------------------------------------------|-----------------------|-------------------------------------------------------------------------------------------------|
| 2015   | Fairthorne et al. | Any psychiatric disorder: Mild–moderate ID: 890 (19.2%)                    | Any psychiatric disorder: 25,818 (9.5%)                                     | Mild–moderate ID: IRR = 1.80, 95% CI [1.5, 2.2]                                          | ICD-9 and ICD-10 codes | Statutory state-based registries of hospital admissions and outpatient mental health appointments |
|        |                   | Severe ID: 52 (16.2%)                                                      |                                                                            | Severe ID: IRR = 0.85, 95% CI [0.4, 1.9]                                                 |                       |                                                                                                  |
| 2016   | Fairthorne et al. | Mothers of children with ID spent 7% more days in hospital compared with mothers of TD children |                                                                            | Any psychiatric disorder: IRR = 1.73 (1.4, 2.1)                                          | ICD-9 and ICD-10 codes | Statutory state-based registries of hospital admissions and outpatient mental health appointments |
|        |                   |                                                                            |                                                                            | Inpatient psychiatric episode treatment: IRR = 1.07 (1.05, 1.10)                         |                       |                                                                                                  |
| 2002   | Olsson & Hwang    | Sense of coherence: ID M = 64.4, SD = 14.5                                 | Sense of coherence: M = 69.1, SD = 11.9                                      | Mothers ID had lower SoC than mothers TD                                                |                       | SoC: 7-point scale, 13 items, e.g. feeling of not being in control, of no meaning to things, of disappointment, higher score indicates higher sense of coherence |
| 2007   | Mugno et al.      | Overall quality of life: M = 63.27, SD = 20.48                             | Overall quality of life: M = 77.98, SD = 13.73                               | F = 9.16, P < .001                                                                      |                       | Overall perceived quality of life; higher score indicates better quality of life                  |
| 2011   | Mirsaleh et al.   | Social dysfunction: M = 7.68, SD = 3.63                                    | Social dysfunction: M = 6.10, SD = 2.64                                     | F = 3.92, P < .001                                                                      |                       | GHQ-28: social dysfunction subscale; higher score indicates worse health                            |
| 2018   | ALAnsari and      | 1. Social dysfunction: M = 1.46, SD = 0.2                                  | 1. Social dysfunction: M = 1.06, SD = 0.28                                   | 1. GHQ-28                                                                             |                       |                                                                                                  |
|        | Jahrami           | 2. Psychological health: M = 70.3, SD = 2.6                                | 2. Psychological health: M = 72.9, SD = 2.2                                  | 2. WHOQOL-BREF                                                                        |                       |                                                                                                  |
|        |                   | 3. GAF mean score: M = 72.61, SD = 2.9                                     | 3. GAF mean score: M = 88.73, SD = 0.60                                     | 3. GAF                                                                                |                       |                                                                                                  |
| 2018   | Hosseinkhan et al.| Emotional adjustment: M = 9.50, SD = 0.96                                 | Emotional adjustment: M = 10.83, SD = 1.07                                  | Difference in at least one of the components: F_{5.90} = 23.64, P = .001               | Bell Adjustment Inventory | 5 components of job matching, home-based, emotional, health and social adjustment; 32 questions with answers of good/I do not know |
|        |                   | Health adjustment: M = 6.16, SD = 0.69                                    | Health adjustment: M = 6.66, SD = 0.75                                      | Emotional adjustment: F_{1.94} = 40.64, P = .001                                       |                       |                                                                                                  |
|        |                   |                                                                            |                                                                            | Health adjustment: F_{1.94} = 11.43, P = .001                                            |                       |                                                                                                  |
Mental ill-health in mothers

Baker (2019) (Fig. 4). Norlin and Broberg (2013) collected data at two time points, but data on stress were available only for time 1. Hosseininik et al. (2018) collected data on four different subscales of the Distress Tolerance Questionnaire, but only results for the tolerance to stress subscale were entered in the meta-analysis. For Ong et al. (1999), only a total score on the Parenting Stress Index was entered in the meta-analysis. The pooled SMD for differences in stress levels between mothers of children with IDs and mothers of typically developing children was 0.82, 95% CI [0.31, 1.33], P = .002.

There was evidence of a high level of heterogeneity between studies, with $I^2 = 90\%$, but the effect size was large, showing that mothers of children with IDs are indeed more stressed than mothers of typically developing children.

**Emotional burden**

Emotional burden was investigated in four studies on mothers of children (Walker et al. 1992; Weiss 2002; Emerson 2003; Lenhard et al. 2005), using a variety of measures, including an unvalidated questionnaire from an unpublished preliminary study (Lenhard et al. 2005).

Two of the papers reported data suitable for meta-analysis on emotional burden/exhaustion (Walker et al. 1992; Weiss 2002). The pooled SMD for differences in levels of emotional burden/exhaustion in mothers of children with IDs and mothers of typically developing children was 1.05, 95% CI [0.21, 1.89], $P = .01$, showing a statistically significant large effect size demonstrating that levels of emotional burden/exhaustion were higher in mothers of children with IDs compared with mothers of other children. However, there was evidence of high statistical heterogeneity between studies, with $I^2 = 79\%$ (Fig. 5).

**Common mental disorders**

Common mental disorders were investigated in seven studies on mothers of children (Emerson 2003; Abasiubong et al. 2006; Mugno et al. 2007; Mirsaleh et al. 2011; Totsika et al. 2011b; AlAnsari and Jahrami 2018; Blacher and Baker 2019). Five used the General Health Questionnaire (Emerson 2003; Abasiubong et al. 2006; Mirsaleh et al. 2011; Totsika et al. 2011b; AlAnsari and Jahrami 2018). All reported
higher level of mental disorders in mothers of people with IDs.

Data were suitable for meta-analysis in four studies, which examined common mental disorders (Mugno et al. 2007; Mirsaleh et al. 2011; AlAnsari and Jahrami 2018; Blacher and Baker 2019) (Fig. 6). The pooled SMD for difference in occurrence of common mental disorders in mothers was 0.94, 95% CI [0.37, 1.52], $P < .001$, showing a large effect size demonstrating that levels of common mental disorders were higher in mothers of children with IDs compared with mothers of typically developing children. However, there was evidence of high statistical heterogeneity between studies, with $I^2 = 87\%$.

**Somatic symptoms**

Somatic symptoms were investigated in four studies on mothers of children (Weiss 2002; Mirsaleh et al. 2011; Long et al. 2015; AlAnsari and Jahrami 2018). Two studies used the somatic symptoms subscale of the General Health Questionnaire (Mirsaleh et al. 2011; AlAnsari and Jahrami 2018) while the remaining studies used a variety of measures. All studies reported more somatic symptoms for mothers of people with IDs apart from one paper (AlAnsari and Jahrami 2018).

Data were suitable for meta-analysis in three studies which investigated somatic symptoms of mental ill-health (Weiss 2002; Mirsaleh et al. 2011; AlAnsari and Jahrami 2018). The pooled SMD for level of somatic symptoms experienced due to mental ill-health by mothers of children with IDs and mothers of typically developing children was 0.15, 95% CI [−0.77, 1.07], $P > .1$, showing a statistically non-significant small effect size demonstrating that levels of these somatic symptoms do not differ greatly between mothers of children with IDs and mothers of typically developing children. There was also evidence of high statistical heterogeneity between studies, with $I^2 = 94\%$ (Fig. 7).

**Psychiatric disorders**

Any psychiatric disorder was investigated in three studies; one on mothers of children (Miller and Keirn 1978) and two on mothers of children and adults (Fairthorne et al. 2015, 2016). Two studies used records in statutory state-based registries of hospital admissions and outpatient mental health appointments (Fairthorne et al. 2015, 2016). All three
studies reported higher levels of psychiatric disorders in mothers of people with IDs, with the exception of hypochondriasis and paranoia subscales investigated by Miller and Keirn (1978), which reported lower rates. Only one study reported data suitable for meta-analysis, so it was not possible to conduct one.

Wellbeing

Six studies investigated wellbeing, using different measures; five on mothers of children (Olsson and Hwang 2002; Mugno et al. 2007; Mirsaleh et al. 2011; AlAnsari and Jahrami 2018; Hosseininik et al. 2018) and one on mothers of adults (Rimmerman and Muraver 2001). Two studies used the General Health Questionnaire to measure social dysfunction (Mirsaleh et al. 2011; AlAnsari and Jahrami 2018) while the remaining studies used a variety of measures to investigate different concepts of wellbeing. One study reported non-significant better wellbeing ratings in mothers of children with IDs compared with mothers of typically developing children (Mugno et al. 2007). Another study found no statistical difference in wellbeing for mothers of adults with IDs compared with typically developing adults (Rimmerman and Muraver 2001). Although all studies reported data suitable for meta-analysis, they investigated very diverse and not easily comparable concepts of wellbeing such as sense of coherence, overall quality of life, social dysfunction, psychological health, emotional and health adjustment, wellbeing, and poor health. For this reason, data on wellbeing were not meta-analysed.

Gaps in evidence base

Only three studies, which met our inclusion criteria, investigated mental ill-health of mothers of adults with IDs, including two by the same author utilising the same statutory state-based registries of hospital admissions and outpatient mental health appointments, which displayed a low risk of bias (Fairthorne et al. 2015, 2016). The third study, which investigated mental ill-health in mothers of adults only, displayed an uncertain risk of bias, including a small and non-representative sample and a report of no statistical differences observed between the studied populations, thus limiting its conclusions (Rimmerman and Muraver 2001). These studies provide much needed insight into the comparative health of mothers of older people with...
IDs, but clearly, more evidence is needed, particularly given the likely increased burden of care arising with ageing-related health difficulties in both mothers and their adult children (Haley and Perkins 2004; Chou et al. 2011). As for all the studies reviewed, findings were not broken down by age group of mothers or their children or stages in the caregiving trajectory.

Discussion

This systematic review extends the previous narrative review of studies without comparison groups (O’Keefe and O’Hara 2008) by providing direct meta-analytic evidence that mental ill-health is more common in mothers of people with IDs than in mothers of typically developing people. This includes higher rates of anxiety, depression, parenting stress, common mental disorders and emotional burden, but not somatic symptoms. However, we must acknowledge the heterogeneity of the included studies, impeding syntheses in this systematic review and in turn, increasing the need for further robust research in this area. Our findings are consistent with previous reports on mothers of children with various disabilities or chronic health conditions (Singer 2006; Bailey et al. 2007; Miodrag et al. 2015) and developmental disabilities (Lee 2013). The ill-health experienced by mothers of people with IDs is highly likely to also impact their children and create a circular vortex of family ill-health, as studies of samples with different disabilities (Witt et al. 2003) and in general population (Plass-Christl et al. 2017) have shown. We also identified considerable gaps in the evidence base, particularly a lack of robust, comparative studies on mothers’ mental ill-health across the caregiving trajectory, which is important given that typically developing children usually become increasingly independent and move on to a much greater extent than children with IDs. Information on mothers’ health is important to identify the pivotal points in the caregiving trajectory when mothers require support to prevent/ameliorate mental ill-health, and to heighten awareness of burden of care for healthcare providers to ensure that families of people with IDs receive the best possible care.

Current evidence and future directions

Although we identified a number of studies, the quality of existing research is limited, particularly regarding sample sizes, sample recruitment, and inclusion criteria. Unlike in a meta-analysis of studies investigating adverse health in parents of children with disabilities and chronic health conditions using the Parenting Stress Index’s Health Sub-domain only (Miodrag et al. 2015), a wide variety of mental ill-health measures and concepts were used in this meta-analysis, to a certain extent impeding synthesis. A meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities also recorded a variety of measures used to study the concept, but this fact did not impede the pooling of the effect sizes (Singer 2006).

As noted above, few studies that met our inclusion criteria investigated mental ill-health in mothers of adults, and none reported findings separately for mothers of different ages or at different stages of the caregiving trajectory. More robust evidence is, therefore, needed in order to further explore and isolate with certainty the primary causes of parental ill-health in order to draw firm conclusions and inform interventions, as well as improving our understanding of the long-term impact of the mental ill-health experienced across the caregiving trajectory on quality of life, suicidal ideation and other health inequalities experienced by mothers of people with IDs. This in turn will allow for informed decision making around support and healthcare service commissioning and provision for families of people with IDs. Given that maternal mental health difficulties are likely to have a substantial impact on family needs and quality of life, both the need for improved support for mothers and the need for further methodologically robust research is clearly indicated.

It is also worth noting that while our review did not focus on the factors, which may have particular impact on mothers’ mental health, these may include a multitude of components such as increased demands on parents’ time because of the increased needs to navigate services and advocate for child’s care (e.g. Haveman et al. 1997), awareness of and experience of stigma (e.g. Ali et al. 2012) or cognition of parenting self-esteem and parental locus of control.
(e.g. Hill and Rose 2009). Some studies have also shown that behaviour problems in populations with IDs may contribute more significantly to parenting stress than the diagnosis of IDs per se (e.g. Baker et al. 2003; Neece and Baker 2008). The aforementioned studies highlight that although the findings consistently show more mental health problems in mothers of people with IDs, the aetiology of these problems may not be solely attributed to the diagnosis of IDs itself but is most likely associated with wider challenges of raising a child with IDs.

Strengths and limitations of this study

Strengths of our review include the prospective registration of the protocol, clear inclusion and exclusion criteria, comprehensive search strategy, including searching multiple databases and no time limits, double rating of paper selection and quality, systematic data extraction, and meta-analyses where possible. However, the review is limited by excluding papers that were not available in English. We also excluded studies with comparison groups with mothers of non-typically developing children, syndrome-specific studies, and studies on samples with developmental delay that did not clearly have IDs. Only 10% of papers were screened by two reviewers (E. R. and K. D.), but there was a perfect agreement between them ($\kappa = 1.00$) and we ensured achieving consistency prior to ceasing double rating. Data extraction was performed only by the first reviewer (E. R.) but was undertaken systematically using a structured database. We were not able to provide details on the age of the children as the majority of the reviewed studies did not report this in detail; hence, we were only able to group reviewed studies by broad developmental stages (i.e. childhood and adulthood). Our aim to determine maternal health across the caregiving trajectory was undermined by the limitations of the evidence base. Methodological, outcome measure and reporting heterogeneity limited the studies that could be included in the meta-analyses and conclusions that could be drawn from them. Indeed, even after restricting the number of synthesised studies, heterogeneity was high in all meta-analyses, and this limitation on reliability of results must be acknowledged. Furthermore, the small number of studies, which reported on the mean, standard deviation and sample size necessary for calculating an unbiased SMD meant that findings from meta-analyses of studies on anxiety, common mental disorders, somatic symptoms of mental ill-health and emotional burden/exhaustion are limited to very few studies. Additionally, included studies were predominantly from high-income countries and included samples of predominantly Caucasian mothers, so there may be unexplored cultural differences in the reviewed studies (e.g. Eisenhower and Blacher 2006; Blacher and Baker 2007; Long et al. 2015). For these reasons, findings from these meta-analyses should be interpreted with caution.

Conclusions

Mothers of people with IDs experience more mental ill-health than mothers of typically developing people, particularly anxiety, depression, parenting stress, common mental disorders and emotional burden. Studies are heterogeneous, and gaps remain, particularly on mothers of adults with IDs, and at different ages and stages of the caregiving trajectory. Healthcare providers require heightened awareness of the high rates of mental ill-health and burden of care placed on mothers of people with IDs and need to provide support for the whole family, not only the person with IDs.

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Conflict of Interest

No conflicts of interest have been declared.

Data Availability Statement

Not applicable, as this study involved a systematic review only.

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**A: Appendix 1: search strategy**

**Search methods for identification of studies**

We searched the following electronic databases for all available years for English language papers: the Cumulative Index to Nursing and Allied Health Literature (CINAHL), Excerpta Medica database (EMBASE), Medical Literature Analysis and Retrieval System Online (MEDLINE) and Psychological Information Database (PsycINFO).

**Electronic searches**

We used the following Ovid MEDLINE search strategy and adapted it as appropriate for other databases:

1. ((mental*) adj3 (handicap* or retard*)).tw.
2. ((disabilit*) adj3 (intellectual)).tw.
3. or/1–2
4. depress*.mp.
5. (mental disorder* or anxi* or anxiety disorder* or (bipolar and related disorder*) or behaviour disorder* or behavior disorder* or delirium or dissociative disorder* or emotional disorder* or mental instability or mood disorder* or affective disorder* or neurosis or personality disorder* or psychosis or psychosomatic disorder* or psychiatric disorder* or stress* or stress related disorder* or stress-related disorder* or paranoid disorder* or psychotic disorder*).mp.
6. psychopatholog*.tw.
7. mental health.tw.
8. mental ill-health.tw.
9. mental illness.tw.
10. (mental adj2 (disorder* or problem* or condition*)).tw.
11. (well-being or wellbeing).tw.
12. or/4–11
13. (mothers or parent).mp.
14. mother*.tw.
15. maternal.tw.
16. (caregiver* or care-giver*).mp.
17. carer*.tw.
18. (mum* or mom*).tw.
19. parent*.tw.
20. or/13–19
21. 3 and 12 and 20
22. Humans/
23. 21 and 22

**Searching other resources**

We checked the reference list of all relevant studies in order to find additional publications which may have been missed by our electronic searches (e.g. unpublished or in-press citations).
### B: Appendix 2: quality assessment scoring for individual CASP items

| CASP item                                                                 | Cummings et al. 1966 | Miller and Keirn 1978 | Harris and McHale 1989 | Walker et al. 1989 | Walker et al. 1992 |
|---------------------------------------------------------------------------|-----------------------|-----------------------|------------------------|---------------------|---------------------|
| 1. Did the study address a clearly focused issue?                         | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 2. Was the cohort recruited in an acceptable way?                         | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 3. Was the exposure accurately measured to minimise bias?                 | Yes                   | Yes                   | Cannot tell            | Yes                 | Yes                 |
| 4. Was the outcome accurately measured to minimise bias?                  | Yes                   | Cannot tell           | Yes                    | Yes                 | Yes                 |
| 5. (a) Have the authors identified all important confounding factors?     | Yes                   | No                    | Cannot tell            | Yes                 | No                  |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | Yes                   | No                    | Cannot tell            | Yes                 | No                  |
| 6. (a) Was the follow up of subjects complete enough?                     | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 6. (b) Was the follow up of subjects long enough?                         | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 7. What are the results of this study?                                   | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 8. How precise are the results?                                          | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 9. Do you believe the results?                                           | Yes                   | Yes                   | Yes                    | Yes                 | Cannot tell         |
| 10. Can the results be applied to the local population?                  | Yes                   | Yes                   | Yes                    | Yes                 | Yes                 |
| 11. Do the results of this study fit with other available evidence?       | Yes                   | Yes                   | Yes                    | Cannot tell         | No                  |
| 12. What are the implications of this study for practice?                | Yes                   | No                    | Yes                    | Yes                 | Yes                 |
| Total score                                                              | A                     | C2.1                  | B2                     | B1                  | C2.1                |

1 Possible scorings for individual items: yes (low risk), cannot tell (unknown risk), no (high risk).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating Bx = uncertain risk of bias for x items, low risk of bias in all other items.

Rating Cx,y = high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.
| CASP item | Andersson 1993 | Ong et al. 1999 | Olsson and Hwang 2001 | Rimmerman and Muraver 2001 | Olsson and Hwang 2002 | Weiss 2002 |
|-----------|----------------|-----------------|----------------------|--------------------------|----------------------|-----------|
| 1. Did the study address a clearly focused issue? | Yes | Yes | Yes | Yes | Yes | Yes |
| 2. Was the cohort recruited in an acceptable way? | Yes | Yes | Yes | Yes | Yes | Yes |
| 3. Was the exposure accurately measured to minimise bias? | Yes | Yes | Yes | Yes | Yes | Yes |
| 4. Was the outcome accurately measured to minimise bias? | Yes | Yes | Yes | Yes | Yes | Yes |
| 5. (a) Have the authors identified all important confounding factors? | No | Cannot tell | Yes | Cannot tell | Yes | Yes |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | No | Cannot tell | Yes | Cannot tell | Yes | Yes |
| 6. (a) Was the follow up of subjects complete enough? | Yes | Yes | Yes | Yes | Yes | Yes |
| 6. (b) Was the follow up of subjects long enough? | Yes | Yes | Yes | Yes | Yes | Yes |
| 7. What are the results of this study? | Yes | Yes | Yes | Yes | Yes | Yes |
| 8. How precise are the results? | Yes | Yes | Yes | Yes | Yes | Yes |
| 9. Do you believe the results? | Yes | Yes | Yes | Yes | Yes | Yes |
| 10. Can the results be applied to the local population? | Yes | Yes | Yes | Yes | Yes | Yes |
| 11. Do the results of this study fit with other available evidence? | Yes | Yes | Yes | Yes | Yes | Yes |
| 12. What are the implications of this study for practice? | Yes | Yes | Yes | Cannot tell | Yes | Yes |

Total score: C2 B1 A B1 B1 A

† Possible scorings for individual items: yes (low risk), cannot tell (unknown risk), no (high risk).

CASP, Critical Appraisal Skills Programme Checklist
Rating A = low risk of bias for all 12 items.
Rating Bx = uncertain risk of bias for x items, low risk of bias in all other items.
Rating Cyx = high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.
| CASP item                           | Emerson 2003 | Eisenhower et al. 2005 | Lenhard et al. 2005 | Abasiubong et al. 2006 | Emerson et al. 2006 |
|------------------------------------|--------------|------------------------|---------------------|------------------------|---------------------|
| 1. Did the study address a clearly focused issue? | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 2. Was the cohort recruited in an acceptable way?   | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 3. Was the exposure accurately measured to minimise bias? | Cannot tell  | Cannot tell            | Cannot tell         | Yes                    | Cannot tell         |
| 4. Was the outcome accurately measured to minimise bias? | Yes          | Yes                    | Cannot tell         | Yes                    | Cannot tell         |
| 5. (a) Have the authors identified all important confounding factors? | No           | Yes                    | Yes                 | Cannot tell            | Yes                 |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | No           | Yes                    | Cannot tell         | Cannot tell            | Yes                 |
| 6. (a) Was the follow up of subjects complete enough? | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 6. (b) Was the follow up of subjects long enough?   | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 7. What are the results of this study?           | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 8. How precise are the results?                 | No           | Yes                    | No                  | No                     | No                  |
| 9. Do you believe the results?                  | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 10. Can the results be applied to the local population? | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 11. Do the results of this study fit with other available evidence? | Yes          | Yes                    | Yes                 | Yes                    | Yes                 |
| 12. What are the implications of this study for practice? | Yes          | Yes                    | Yes                 | Yes                    | No                  |
| Total score                                      | C2.1         | B1                     | C1.3                | C1.1                   | C2.2                |

†Possible scorings for individual items: yes (low risk), cannot tell (unknown risk), no (high risk).

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Rating Bx = uncertain risk of bias for x items, low risk of bias in all other items.
Rating Cx,y = high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.
### CASP item†

| CASP item                                                                 | Olsson and Hwang 2006 | Mugno et al. 2007 | Neece and Baker 2008 | Olsson and Hwang 2008 | Gupta and Kaur 2010 |
|---------------------------------------------------------------------------|-----------------------|-------------------|----------------------|-----------------------|---------------------|
| 1. Did the study address a clearly focused issue?                         | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 2. Was the cohort recruited in an acceptable way?                         | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 3. Was the exposure accurately measured to minimise bias?                 | Yes                   | Yes               | Yes                  | Cannot tell           | Yes                 |
| 4. Was the outcome accurately measured to minimise bias?                  | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 5. (a) Have the authors identified all important confounding factors?     | Cannot tell           | Yes               | Yes                  | Yes                   | No                  |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | Cannot tell           | Yes               | Yes                  | Yes                   | No                  |
| 6. (a) Was the follow up of subjects complete enough?                     | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 6. (b) Was the follow up of subjects long enough?                         | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 7. What are the results of this study?                                    | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 8. How precise are the results?                                          | Yes                   | Yes               | Cannot tell           | Yes                   | No                  |
| 9. Do you believe the results?                                            | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 10. Can the results be applied to the local population?                   | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 11. Do the results of this study fit with other available evidence?       | Yes                   | Yes               | Yes                  | Yes                   | Yes                 |
| 12. What are the implications of this study for practice?                 | Cannot tell           | Yes               | Yes                  | Cannot tell           | No                  |
| Total score                                                               | B2                    | A                  | B1                   | B2                    | C3                  |

†Possible scorings for individual items: yes (low risk), cannot tell (unknown risk), no (high risk).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating Bx = uncertain risk of bias for x items, low risk of bias in all other items.

Rating Cy,x = high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.
E. Rydzewska et al. • Mental ill-health in mothers

| CASP item† | Mirsaleh et al. 2011 | Totsika et al. 2011a | Totsika et al. 2011b | Kilic et al. 2013 | Norlin and Broberg 2013 |
|------------|----------------------|----------------------|----------------------|------------------|------------------------|
| 1. Did the study address a clearly focused issue? | Yes | Yes | Yes | Yes | Yes |
| 2. Was the cohort recruited in an acceptable way? | Yes | Yes | Yes | Yes | Yes |
| 3. Was the exposure accurately measured to minimise bias? | Yes | Yes | Cannot tell | Cannot tell | Yes |
| 4. Was the outcome accurately measured to minimise bias? | Yes | Yes | Yes | Yes | Yes |
| 5. (a) Have the authors identified all important confounding factors? | No | Cannot tell | Cannot tell | No | Yes |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | No | Cannot tell | Cannot tell | No | Yes |
| 6. (a) Was the follow up of subjects complete enough? | Yes | Yes | Yes | Yes | Yes |
| 6. (b) Was the follow up of subjects long enough? | Yes | Yes | Yes | Yes | Yes |
| 7. What are the results of this study? | Yes | Yes | Yes | Yes | Yes |
| 8. How precise are the results? | Yes | Yes | Yes | Yes | Yes |
| 9. Do you believe the results? | Yes | Yes | Yes | Yes | Yes |
| 10. Can the results be applied to the local population? | Yes | Yes | Yes | Yes | Yes |
| 11. Do the results of this study fit with other available evidence? | Yes | Yes | Yes | Yes | Yes |
| 12. What are the implications of this study for practice? | No | Yes | Yes | Yes | Yes |
| Total score | C2 | B1 | B2 | C1.1 | A |

†Possible scorings for individual items: yes (low risk), cannot tell (unknown risk), no (high risk).

CASP, Critical Appraisal Skills Programme Checklist.
Rating A = low risk of bias for all 12 items.
Rating B = uncertain risk of bias for all 12 items.
Rating C = high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.

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### CASP item

| CASP item                                                                 | Fairthorne et al. 2015 | Long et al. 2015 | Faithorne et al. 2016 | AlAnsari and Jahrami 2018 | Hosseininik et al. 2018 | Blacher and Baker 2019 |
|--------------------------------------------------------------------------|------------------------|-----------------|-----------------------|---------------------------|-------------------------|------------------------|
| 1. Did the study address a clearly focused issue?                        | Yes                    | Yes             | Yes                   | Yes                       | Yes                     | Yes                    |
| 2. Was the cohort recruited in an acceptable way?                         | Yes                    | Yes             | No                    | Yes                       | Yes                     | Yes                    |
| 3. Was the exposure accurately measured to minimise bias?                 | Yes                    | Yes             | Yes                   | Cannot tell               | Yes                     | Yes                    |
| 4. Was the outcome accurately measured to minimise bias?                  | Yes                    | Yes             | Yes                   | Yes                       | Yes                     | Yes                    |
| 5. (a) Have the authors identified all important confounding factors?     | Yes                    | Yes             | Yes                   | No                        | Yes                     | Yes                    |
| 5. (b) Have they taken account of the confounding factors in the design and/or analysis? | Yes                    | Yes             | Cannot tell           | No                        | Yes                     | Yes                    |
| 6. (a) Was the follow up of subjects complete enough?                     | Yes                    | Yes             | Yes                   | Yes                       | Yes                     | Yes                    |
| 6. (b) Was the follow up of subjects long enough?                         | Yes                    | Yes             | Yes                   | Yes                       | Yes                     | Yes                    |
| 7. What are the results of this study?                                   | Yes                    | Cannot tell     | Yes                   | No                        | Yes                     | Yes                    |
| 8. How precise are the results?                                          | Yes                    | Cannot tell     | Yes                   | Yes                       | No                      | Yes                    |
| 9. Do you believe the results?                                            | Yes                    | Yes             | No                    | Yes                       | No                      | Yes                    |
| 10. Can the results be applied to the local population?                   | Yes                    | Yes             | Yes                   | Cannot tell               | Yes                     | Yes                    |
| 11. Do the results of this study fit with other available evidence?       | Yes                    | Yes             | Yes                   | Yes                       | Yes                     | Yes                    |
| 12. What are the implications of this study for practice?                 | Yes                    | Yes             | No                    | No                        | Yes                     | Yes                    |

**Total score**
- **A**: low risk of bias for all 12 items.
- **B**: uncertain risk of bias for x items, low risk of bias in all other items.
- **C**: high risk of bias in y items, uncertain risk of bias in x items, low risk of bias in all other items.

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