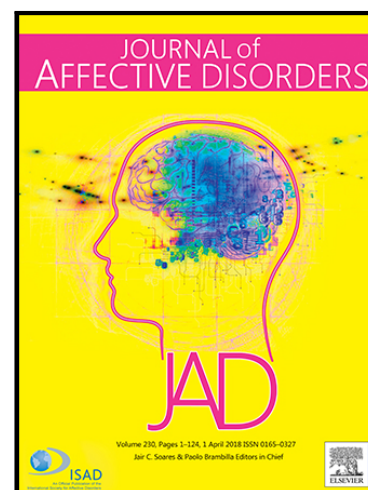


Accepted Manuscript

Prevalence of Anxiety and Depressive Disorders among Youth with Intellectual Disabilities: A Systematic Review and Meta-Analysis

Christophe Maïano , Sylvain Coutu , Danielle Tracey ,
Stéphane Bouchard , Geneviève Lepage , Alexandre J.S. Morin ,
Grégory Moullec

PII: S0165-0327(17)32571-5
DOI: [10.1016/j.jad.2018.04.029](https://doi.org/10.1016/j.jad.2018.04.029)
Reference: JAD 9683



To appear in: *Journal of Affective Disorders*

Received date: 12 December 2017
Revised date: 4 March 2018
Accepted date: 4 April 2018

Please cite this article as: Christophe Maïano , Sylvain Coutu , Danielle Tracey , Stéphane Bouchard , Geneviève Lepage , Alexandre J.S. Morin , Grégory Moullec , Prevalence of Anxiety and Depressive Disorders among Youth with Intellectual Disabilities: A Systematic Review and Meta-Analysis, *Journal of Affective Disorders* (2018), doi: [10.1016/j.jad.2018.04.029](https://doi.org/10.1016/j.jad.2018.04.029)

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

Highlights

- This meta-analysis focuses on anxiety and depressive disorders among youth with ID
- Pooled prevalence of anxiety and depressive disorders were 5.4% and 2.8%
- Pooled prevalence for subtypes of anxiety disorders ranged from 0.2%-11.5%
- Pooled prevalence for subtypes of depressive disorders ranged from 2.5%-3.4%
- Pooled prevalence significantly differed as a function of studies characteristics

Running title: Anxiety, Depression, and Intellectual Disabilities

Prevalence of Anxiety and Depressive Disorders among Youth with Intellectual Disabilities: A Systematic
Review and Meta-Analysis

Christophe Maïano^{1,2,3}, Sylvain Coutu², Danielle Tracey⁴, Stéphane Bouchard^{1,2}, Geneviève Lepage²,
Alexandre J. S. Morin³, Grégory Moullec^{2,5,6}

¹ Cyberpsychology Laboratory, Department of Psychoeducation and Psychology, Université du Québec en Outaouais (UQO), Gatineau, Canada.

² Department of Psychoeducation and Psychology, Université du Québec en Outaouais (UQO), Saint-Jérôme, Canada.

³ Substantive-Methodological Synergy Research Laboratory, Department of Psychology, Concordia University, Montréal, Canada.

⁴ School of Education, Western Sydney University, Sydney, Australia

⁵ School of Public Health, Department of Social and Preventive Medicine, Université de Montréal, Montréal, Canada

⁶ Research Center, Centre intégré universitaire de santé et de services sociaux (CIUSSS) du Nord-de-l'Île-de-Montréal, Montréal, Canada

Corresponding author: Christophe Maïano, Université du Québec en Outaouais, Campus de Saint-Jérôme, Département de Psychoéducation et de Psychologie, 5 rue Saint-Joseph, Saint-Jérôme, Québec, J7Z 0B7, email: christophe.maïano@uqo.ca

Abstract

Background. The purpose of this meta-analytic study was to determine the pooled prevalence estimates of anxiety and depressive disorders among children and adolescents with intellectual disabilities (ID) and to assess the extent to which these pooled prevalence rates differed according to studies' characteristics.

Method. A systematic literature search was performed in nine databases and 20 studies, published between 1975 and 2015, met the inclusion criteria.

Results. The resulting pooled prevalence estimates of combined subtypes of anxiety and depressive disorders were respectively (a) 5.4% and 2.8% across samples; (b) 1.2% and 0.03% among children; and (c) 7.9% and 1.4% among adolescents. Pooled prevalence estimates for specific subtypes of anxiety disorders ranged from (a) 0.2% to 11.5% across samples; (b) 0.7% to 17.6% among children; and (c) 0.6% to 19.8% among adolescents. Pooled prevalence estimates of dysthymic disorder and major depressive disorder were respectively (a) 3.4% and 2.5% across samples; (b) 2.1% and 3.2% among children; and (c) 6.9% and 5.7% among adolescents. Finally, subgroup analyses showed significant variations in the pooled prevalence estimates of combined subtypes of anxiety disorders, obsessive-compulsive disorder, and generalized anxiety disorder; and combined subtypes of depressive disorders.

Limitations. The present findings of this meta-analysis should be interpreted with caution given several limitations related to the characteristics of the populations, diagnosis method and sampling method.

Conclusion. Findings provide recommendations for future studies investigating psychological disorders among youth with ID, as well as how clinicians and policy makers can improve diagnostic practices and support for youth with ID.

Keywords: diagnostic criteria, informant, intellectual disability level, moderation, psychological disorders

ACCEPTED MANUSCRIPT

1. Introduction

The last two decades have witnessed a growing research interest in the prevalence of psychological disorders among youth with intellectual disabilities (ID) and their results have been summarized in a few systematic reviews¹ (e.g., Einfeld, Ellis, & Emerson, 2011; Hudson & Chan, 2002; Oeseburg, Dijkstra, Groothoff, Reijneveld, & Jansen, 2011; Whitaker & Read, 2006). Overall, these reviews reveal that a large proportion of youth with ID present with at least one psychological disorder, although exact prevalence rates remain poorly documented (Oeseburg et al., 2011). For example, Einfeld et al. (2011), and Whitaker and Read (2006) reported that between 30%-50% of children and 24%-54% of adolescents with ID experience at least one psychological disorder. Einfeld et al. (2011) further showed that this rate was 2.8 to 4.5 times greater for youth with ID than the rates obtained among typically developing (TD) youth.

Although the proportion of youth with ID experiencing psychological disorders, in general, has been addressed within the literature, reviews focusing on prevalence estimated for specific psychological disorders in this population remain limited. In particular, no systematic review or meta-analysis have yet provided a summary of findings, or analysis of findings, from prevalence studies focusing specifically on depressive disorders among youth with ID or reported pooled prevalence estimates of depressive disorders among this population. Additionally, although Reardon, Gray, and Melvin (2015) recently summarized studies on the prevalence of anxiety disorders among youth with ID, their initial effort presents several shortcomings that need to be addressed. First, the pooled prevalence of specific and combined subtypes of anxiety disorders have not been estimated, notably those of *anxiety-related disorders* (as defined in previous diagnostic classifications), such as obsessive-compulsive or posttraumatic stress disorders. Second, Reardon et al. (2015) did not examine whether the prevalence estimates of specific and combined subtypes of anxiety disorders differ according to the youth's

characteristics, such as age (children vs. adolescents-young adults), sex (boys vs. girls), ID level (e.g., mild vs. moderate or severe), and geographic location (e.g., North America vs. Europe). Third, Reardon et al. (2015) did not investigate the potential moderating role played by the specific source of information used to assess the presence of anxiety disorders (e.g., medical records, interview, clinical judgment, or multiple assessments).

The specific focus of the present article on anxiety and depressive disorders is highly relevant. Firstly, longitudinal studies with TD youth have established that anxiety and depressive disorders in childhood and adolescence serve as significant predictors of future mood disorders (Roza, Hofstra, van der Ende, & Verhulst, 2003) and major depression in adulthood (Reinherz, Paradis, Giaconia, Stashwick, & Fitzmaurice, 2003). Given their role as precursors for adulthood wellbeing, research must seek to determine the specific prevalence of anxiety and depressive disorders for youth with ID and whether these disorders are prevalent at higher or lower levels than in TD youth in order to guide the preventative work of policy makers, researchers, and practitioners. Secondly, although previous systematic reviews investigating a wider range of psychological disorders in general are helpful (e.g. Einfeld et al., 2011; Oeseburg et al., 2011), a specific focus on anxiety and depression affords a more thorough and nuanced analysis of both the potential sample and study characteristics that may be driving disputed prevalence rates for youth with ID. Providing precise and up to date information is critical to inform future research and practice in the identification and management of anxiety and depression disorders for youth with ID.

With the present systematic review and meta-analysis we intend to address the shortcomings of previous research. More specifically, the first objective was to estimate the pooled prevalence rates of anxiety and depressive disorders (specific and combined subtypes) for all of the samples in this review, as well as separately for samples of children and adolescents. The second objective was to examine

whether the pooled prevalence rates of anxiety and depressive disorders differ as a function of specific characteristics of the studies.

2. Method

This systematic review and meta-analysis was performed following guidelines from the Preferred Reporting Items for Systematic reviews and Meta-Analyses statement (PRISMA; Liberati et al., 2009) and the Meta-Analysis Of Observational Studies in Epidemiology statement (MOOSE; Stroup et al., 2000).

2.1 Information Sources and Search Strategy

Potentially relevant studies were identified through a systematic and simultaneous electronic search in Academic Search Complete, CINAHL Plus with Full-Text, Education Sources, ERIC, Medline with Full-Text, Psychology and Behavioral Sciences Collection, and SocINDEX via the EBSCO database. In addition, a systematic electronic search was conducted separately in the PsycARTICLES (including PsycINFO) and Scopus databases. No year restriction was imposed in the electronic search and the last updated search was performed on the 26th of November 2016. Studies were identified in the aforementioned databases using the following three groups of search terms: (1) "intellectual* dis*" OR "mental* retard*" OR "developmental dis*" OR "developmental del*"; AND (2) anxiet* OR anxious OR depress* OR "affect* dis*"; AND (3) child* OR adolescen* OR student* OR youth* OR paediatric* OR pediatric*. The search strategy used in Scopus database is presented in the online supplement (see the section S1). More precisely, these groups were combined and researched in the title, abstract and keywords of the studies published by the journals indexed in the searched databases. Finally, a manual search was also conducted in: (a) the reference lists of the articles included in the meta-analysis and in the manuscripts citing these articles; and (b) in previous literature reviews on anxiety (Reardon et al., 2015) and psychological disorders (Borthwick-Duffy, 1994; Einfeld et al., 2011; Hemmings et al., 2013; Hudson & Chan, 2002; Oeseburg et al., 2011; Whitaker & Read, 2006; Yoo et al., 2012) among people

with ID.

2.2 Inclusion Criteria

Only the studies meeting the following inclusion criteria were included in the meta-analysis. First, participants had to present mild, moderate, severe, or profound ID² (or a developmental delay) of known (i.e., autism spectrum disorder, Down syndrome, Fragile X syndrome, Williams syndrome, Prader-Willi, etc.) or unknown etiology. However, studies exclusively focusing on participants with ID of a specific known etiology were not included. The rationale for excluding these studies is that: (1) we could not be confident that all studies limited to these subpopulations would be identified as they often do not focus on their ID levels as does the current meta-analysis; (2) the prevalence of anxiety or depression disorders has already been quantified for these subpopulations (e.g., Royston, Howlin, Waite, & Oliver, 2017; van Steensel, Bögels, & Peerin, 2011); and (3) the phenotype of people from these subpopulations can heighten their vulnerability to anxiety or depressive disorders, and thus bias prevalence estimates associated with more generic forms of ID (e.g., Royston et al., 2017; van Steensel et al., 2011). Finally, studies which included a range of disabilities within the sample were considered to be eligible if the data on the relevant outcomes were available for participants with ID.

Second, the participants with ID included in the studies had to be infants (0-3 years), children (4-11 years), or adolescents-young adults (12-22 years). Mixed samples of adolescents and adults with ID were included if the data on the relevant outcomes were presented for the participants aged 22 years and lower. Studies exclusively focusing on samples of adults with ID were excluded.

Third, studies were considered to be eligible only if one of their main objective was to determine the prevalence of anxiety (e.g., combined subtypes of anxiety disorder³, agoraphobia, generalized anxiety disorder, obsessive-compulsive disorder) and/or depressive disorders (i.e., combined subtypes of depressive disorders⁴, dysthymic disorder, major depressive disorder) among youth with ID. Therefore,

studies including participants with a diagnosed anxiety disorder and/or depressive disorder aiming to compare their clinical features with those from non-clinical participants, or to validate screening instruments, were not included. In addition, when the same sample (or a part of a sample) was used in several publications, the most recent or the largest sample was included. In this situation, outcomes of interest not reported in the selected publication were also included from the other publication.

Fourth, for studies to be included, anxiety and/or depressive disorders had to be diagnosed by qualified professionals (e.g., physician, psychiatrist, psychologist, pediatrician) and/or obtained by the research team through a structured diagnostic interview. Therefore, studies focusing on specific symptoms or using screening cut-off scores (e.g., Child Behavior Checklist, Developmental Behavior Checklist) were excluded. Indeed, the focus here was on the prevalence of psychiatric diagnoses, rather than on either the clinical severity of symptoms or the presence of participants “at risk” of meeting clinical criteria diagnoses. In addition, the diagnosis performed by qualified professionals requires a discussion with the target participant and/or informant as well as an assessment of the severity and discomfort associated with the assessed symptoms, which is typically lacking as part of screening severity procedures relying on participant and/or informant reports.

Finally, we only included studies relying on cohort (only the first or initial measure was considered), cross-sectional or case-control design written in English and published or in-press in a peer-reviewed journal. Case studies, book chapters, conference proceedings, and non-original studies (i.e., comments, reviews, theoretical papers) were excluded.

2.3 Selection of the Relevant Studies

As recommended by the PRISMA Statement (Liberati et al., 2009), the eligibility of the relevant studies was determined based on the examination of the titles-abstracts and full texts. First, the relevance of the titles-abstracts of the studies were independently screened by three authors (first,

second, and last). Then, the full texts of the studies selected based on their titles-abstracts were also independently screened by the same three authors to assess their eligibility. At each step, discrepancies between authors were resolved by discussion until an agreement was reached.

2.4 Data Extraction

The same three authors independently extracted the information and data presented in the full text articles of studies included in the meta-analysis. The following information was extracted: (a) location (country); (b) design (cohort, cross-sectional, case control); (c) recruitment setting (e.g., service agencies, special school, child psychiatric unit); (d) type of samples (children, adolescents, mixed); (e) characteristics of samples with ID (i.e., sample size, percentage of boys, age range, and ID level); (f) presence/absence (yes/no) and sample size of TD participants; (g) diagnostic information (i.e., informant, method, and criteria); (h) types of anxiety disorders (e.g., combined subtypes, agoraphobia without panic disorder, generalized anxiety disorder, any anxiety disorders); (i) types of depressive disorders (e.g., combined subtypes, depressive disorders, dysthymic disorder, major depressive disorder); (j) prevalence estimates (i.e., the percentage and the sample size or the frequencies) of anxiety and depressive disorders. The information and the data extracted were reviewed and discrepancies were resolved by discussion.

2.5 Quality Assessment of the Studies Included in the Meta-Analysis

An adapted version of the Methodological evaluation of Observational REsearch (MORE) checklist for observational studies of incidence or prevalence of chronic diseases was used to assess the quality of reporting of studies (Shamliyan et al., 2013). The scoring regarding the external (i.e., sampling method, estimation of sampling bias, sampling bias in the analysis) and internal validity (i.e., source, definition and measurements of anxiety/depression) was completed independently by the first and last

authors. Their results were then reviewed by the two authors, and remaining disagreements were resolved by the second author.

2.6 Statistical Analysis

All the analyses were performed using the version 2.2.064 of the Comprehensive Meta-Analysis (CMA) software developed by Borenstein, Hedges, Higgins, and Rothstein (2005). Given the heterogeneity of the studies included in the meta-analysis (participants' characteristics, diagnostic method, sample size, etc.), a random effects model was used to estimate the pooled or weighted prevalence of anxiety and depressive disorders. First, the pooled or weighted prevalence rates of anxiety and depressive disorders were estimated including all the relevant studies. Second, the pooled or weighted prevalence rates of anxiety and depressive disorders were separately estimated for studies including children or adolescents. The forest plots of these pooled prevalence estimates were graphed using the Microsoft Excel spreadsheet developed by Neyeloff, Fuchs, and Moreira (2012). The heterogeneity of the pooled prevalence estimates was examined using Cochran's (1950) Q test and Higgins, Thompson, Deeks, and Altman's (2003) I^2 statistic. Finally, several statistical tests provided by the CMA software were used (Begg and Mazundar's rank correlation test, 1994; Duval and Tweedie's "trim and fill" test, 2000; Egger's test of the intercept, 1997) to assess potential publication bias in the pooled prevalence estimates of anxiety and depressive disorders.

Moderation analyses were examined using a mixed effect model. We performed a series of pre-specified subgroup analyses for the four following variables: (a) ID level (borderline, mild, moderate, severe, profound, unspecified); (b) geographic regions as defined by the World Health Organization (Europe, North America, South America); (c) diagnostic method (interview, medical records, multiple assessments); (d) diagnostic criteria (the diagnostic and statistical manual of mental disorders [DSM]: second, third, third-revised, fourth, and text revision of the fourth edition; and tenth edition of the

international statistical classification of diseases and related health problems [ICD-10]); and (e) informant (e.g., caregiver, caregiver and parents, parents, parents and youth). No moderation analyses were performed when only one study was available in a pre-specified subgroup.

3. Results

3.1 Study Selection

The electronic and manual search identified a total of 10,671 relevant articles, which fell to 7,786 when duplicates were removed. A total of 7,673 articles were excluded based on the analysis of their title and abstract. Then, the full text of the remaining 113 articles (see section S2 in the online supplements for the full references of these studies) were assessed and 92 were excluded for reasons presented in Figure 1. Therefore 21 studies, detailed in Tables 1 and 2 and published between 1975 and 2015, met the inclusion criteria and were included in this meta-analysis.

3.2 Study Characteristics

Design and Sample Characteristics. As illustrated in Table 1, most of the studies were conducted in North America ($n = 9$) and Europe ($n = 10$). More than two third of the studies (15/21) had a cross-sectional design, and only four had included a TD sample for comparative analyses. Overall, a total of 57,971 youth with ID ($M = 2,760$; range = 30 to 43,738) participated in these studies, and 12/21 studies (57.1%) recruited participants from service agencies or schools. Additionally, the majority of studies (13/21, 61.9%) included participants with borderline-mild to severe-profound ID. In addition, 3/21 studies (14.3%) focused on children (4–11 years), 5/21 (23.8%) on adolescents-young adults (12–21 years), and 13 of the 21 (61.9%) focused on mixed-age sample (e.g., infants to adolescents-young adults, children and adolescents). Participants from these studies were mostly boys ($M = 61%$, range = 53% to 70%).

Diagnostic Information. As presented in Table 2, the information used to diagnose anxiety or depressive disorders was mostly obtained from multiple respondents ($n = 6$; i.e., parents, youth, and/or caregivers) or from parents only ($n = 7$). Additionally, diagnoses have been obtained using multiple assessments (33.3%), medical records (33.3%), and interviews (33.3%). Finally, these diagnoses were mostly determined based on DSM criteria (57.1%), followed by the ICD-10 criteria (33.3%).

Types of Anxiety and Depressive Disorders. As illustrated in Table 2, 19/21 (90.5%) studies reported the prevalence of anxiety disorders, and one third focused on diagnostic subtypes such as generalized anxiety disorder, obsessive-compulsive disorder, posttraumatic stress disorder, separation anxiety disorder, etc. Additionally, 15/21 studies (71.4%) reported the prevalence of depressive disorders, and one third focused on diagnostic subtypes such as dysthymic disorder and major depressive disorder.

3.3 Prevalence Estimates: Anxiety Disorders

All Samples. Prevalence estimates of combined subtypes of anxiety disorders were reported in 18 studies (see Figure 2a for the forest plot). Prevalence estimates are reported in Table 3. Across these studies, the pooled prevalence estimate was 5.4% (95% CI = 2.5–11.5), with a very high level of heterogeneity. No evidence of publication bias was noted by most of the tests, except for the Duvall and Tweedie's trim and fill test which revealed that one study was missing on the left of the funnel plot. When this study was imputed to obtain a symmetrical funnel plot, the pooled prevalence estimate became 4.7% (95% CI = 2.1–10).

Specific subtypes. Ten specific subtypes of anxiety disorders were reported in the studies (see Figures S1a-f and Table S1 in the online supplements for forest plots and references). Their prevalence estimates ranged from 0.2% for panic disorder with agoraphobia to 11.5% for specific phobia (Table 3). No evidence of publication bias was found for most of the specific subtypes, except for specific phobia

for which the Duvall and Tweedie's trim and fill revealed that one study was missing. The imputation of this missing study gave a pooled prevalence estimate of 10.2% (95% CI = 4.1–23.2) for specific phobia.

Children Samples. Prevalence estimates of combined subtypes of anxiety disorders among children were reported in three studies (Figure 2b). The pooled prevalence estimate (Table 3) was 1.2% (95% CI = 0–49.1), with a very high percentage of heterogeneity. The Duvall and Tweedie's trim and fill revealed that two studies were missing. The imputation of these missing studies gave a pooled prevalence estimate of 0.1% (95% CI = 0–5.5).

Specific subtypes. Only one study (Table S1) reported prevalence estimates for three specific subtypes of anxiety disorders (generalized anxiety disorders, separation anxiety disorder, and social phobia). These prevalence estimates range from 0.7% for generalized anxiety disorders to 18% for separation anxiety disorder (Table 3).

Adolescent Samples. Prevalence estimates of combined subtypes of anxiety disorders were reported in five studies (Figure 2c). The pooled prevalence estimate was 7.9% (95% CI = 0.6–54.6), with a very high percentage of heterogeneity (Table 3). The Duvall and Tweedie's trim and fill revealed that three studies were missing to obtain a symmetrical funnel plot. The imputation of these missing studies gave a pooled prevalence estimate of 0.5% (95% CI = 0–7.8).

Specific subtypes. Seven specific subtypes of anxiety disorders were reported in the studies (Table S1). In these studies, prevalence estimates ranged from 0.6% for panic disorder to 19.8% for specific phobia (Table 3). Given the very few number of studies per anxiety subtype, no publication bias analyses were performed.

3.4 Prevalence Estimates: Depressive Disorders

All Samples. Prevalence estimates of combined subtypes of depressive disorders were reported in 10 studies (Figure 3a). Prevalence estimates are reported in Table 4. Across these studies, the pooled prevalence estimate was 2.8% (95% CI = 1.1–6.7), with a very high percentage of heterogeneity. No evidence of publication bias was noted by most of the tests, except for the Duvall and Tweedie's trim and fill test revealing that three studies were missing. When these studies were imputed, the pooled prevalence estimate became 1.6% (95% CI = 0.6–3.7).

Specific subtypes. Two specific subtypes of depressive disorders were reported in the selected studies (see Figures S2a-b and Table S2 in the online supplements for forest plots and references). Pooled prevalence estimates were 3.4% for dysthymic disorder and 2.5% for major depressive disorder (Table 4). No publication bias was found.

Children Samples. Combined subtypes of depressive disorders were considered in only one study (Table S2), which reported a prevalence estimate of 0.03% (95% CI = 0.0–0.1).

Specific subtypes. Two specific subtypes of depressive disorders were considered in the studies (Table S2), and their pooled prevalence estimates were 2.1% for dysthymic disorder and 3.2% for major depressive disorder. Given the very few number of studies per depressive subtype, no publication bias analyses were performed.

Adolescent Samples. Prevalence estimates of combined subtypes of depressive disorders were reported in three studies (Figure 3b). The pooled prevalence estimate was 1.4% (95% CI = 0.4–5.1), with a very high percentage of heterogeneity (Table 4). The Duvall and Tweedie's trim and fill test revealed that two studies were missing. The imputation of these studies provided a pooled prevalence estimate of 0.5% (95% CI = 0.2–1.6).

Specific subtypes. Two specific subtypes of depressive disorders were considered in the studies (Table S2), and their pooled prevalence estimates were 6.9% for dysthymic disorder and 5.8% for major depressive disorder (Table 4). Given the very few number of studies per depressive subtype, no publication bias analyses were performed.

3.5 Moderation Analyses

Results from the moderation analyses are detailed in Tables S3-S6 in the online supplements. Given the low number of studies comprising samples of children and adolescents only, the analyses were only performed across all samples (children, adolescents, and mixed), rather than on age-specific samples.

Anxiety Disorders. The results showed significant differences in pooled prevalence estimates of combined subtypes of anxiety disorders and obsessive-compulsive disorder according to the diagnostic method that was used (Table S3). Pairwise comparisons showed that the prevalence estimate of combined subtypes of anxiety disorders was significantly higher when participants were diagnosed by interview (22.5%) than by multiple assessments (5.3%) or medical records (1.9%). In addition, the pooled prevalence estimate of obsessive-compulsive disorder among youth with ID was significantly higher when participants were diagnosed by interview (7.3%) than by multiple assessments (0.4%).

The results also showed significant differences in the pooled prevalence estimates of combined subtypes of anxiety disorders, obsessive-compulsive disorder, and generalized anxiety disorder as a function of the diagnostic criteria that were used (Table S3). Pairwise comparisons showed higher rates of combined subtypes of anxiety disorders when participants were diagnosed using the DSM-IV (21.4%) rather than the DSM-III (6.5%), ICD-10 (4.4%), or DSM-III-R (0.6%) criteria. The prevalence of obsessive-compulsive disorders was also higher when participants were diagnosed using the DSM-IV-TR (12%)

rather than the ICD-10 (0.4%) criteria. However, prevalence of generalized anxiety disorder was higher when participants were diagnosed using the ICD-10 (2.3%) rather than DSM-IV (0.3%) criteria.

Finally, results showed a significant difference in pooled prevalence estimates of combined subtypes of anxiety disorders according to the informant (Table S3). More specifically, rates of combined subtypes of anxiety disorders were higher when the informant was the parents (15%) rather than the parents and youth (4.4%). Finally, no significant variations were found as a function of ID level and geographic location.

Depressive Disorders. The results showed significant differences in prevalence of combined subtypes of depressive disorders according to the ID level of the participants (Table S5). Pairwise comparisons showed that prevalence estimates were significantly higher among youth with a borderline ID (17.1%) than among those with a moderate (3.5%), severe (2.6%), or unspecified ID (3.4%). No significant variations were found as a function of geographic location, diagnostic method and criteria, and informant.

3.6 Quality Assessment of the Studies

Table 5 reports the quality ratings of studies based on the adapted MORE's criteria (Shamliyan et al., 2013). Only six studies (6/21, 28.6%) used a population-based sample raising concerns about the representativeness of the results obtained in terms of prevalence. Moreover, most of the studies (17/21, 80.9%) failed to report the response rate (meaning that we cannot estimate the sampling bias) and the subject flow (20/21, 95.2%). However, nine studies performed subgroup analyses (mostly according to age and/or sex subgroups).

Regarding the internal validity, only seven studies (7/21, 33.3%) reported and justified the reference period (i.e., current or lifetime) for the diagnosis of anxiety and depression. Moreover, the majority of

the studies reported information about the source of the measure (15/21, 71.4%) and whether the outcomes were measured for the purpose of the study (11/21, 52.4%). Finally, most of the studies did not document the validity and reliability of the instruments used to measure anxiety and depression (16/21, 76.2%), nor did they report information about the precision of estimate (17/21, 80.9%) or adjusted estimates (19/21, 90.5%).

4. Discussion

4.1 Prevalence Estimates of Anxiety and Depressive Disorders

The first objective of this review and meta-analysis was to provide a synthesis of the empirical studies examining the prevalence of anxiety and depressive disorders among children and adolescents with ID, and then to estimate the pooled or weighted prevalence of these disorders among these populations combined and separately.

Anxiety Disorders. The findings revealed a pooled prevalence estimate for combined subtypes of anxiety disorders of 5.4% across samples, with a large heterogeneity (ranging from 0.1% to 40%). This prevalence estimate is slightly lower than those found in recent meta-analyses of samples of TD youth, where the pooled prevalence of combined subtypes of anxiety disorders were 6% (Baxter et al., 2013), 6.5% (Polanczyk et al., 2015), and 10.2% (Costello et al., 2011). In addition, the current meta-analysis showed that prevalence of combined subtypes of anxiety disorders were six times more important among adolescents with ID (7.9%; ranging from 0.2% to 40%) than among children (1.2%; 0.1% to 17.6%). This result is not in line with Costello et al.'s (2011) meta-analysis that found comparable prevalence rates in TD children (12%) and adolescents (11%). Finally, 10 specific subtypes of anxiety disorders were examined in the reviewed studies. Findings showed that (a) specific phobia (11.5%) was highly prevalent across all samples; (b) children with ID suffer more frequently from separation anxiety disorders (17.6%); and (c) adolescents with ID suffer more frequently from specific phobia (19.8%),

obsessive-compulsive disorder (12%), and social phobia (8%). These findings are consistent with those found by Costello et al. (2011) among TD youth.

Depressive Disorders. Our results revealed a pooled prevalence estimate for combined subtypes of depressive disorders of 2.8% (range = 0.1%-14%) across samples. This prevalence estimate matches the 2.6% found by Polanczyk et al. (2015) in their meta-analysis among TD youth. Additionally, even if these prevalence estimates remain low, it is interesting to note that combined subtypes of depressive disorders are more frequent in adolescents with ID (1.4%; range = 0.5%-2.7%) than children (0.3%). This greater risk of depressive disorders for adolescents is well documented among TD youth (e.g., Birmaher et al., 1996). More specifically, pooled prevalence estimates were higher for major depressive disorders (3.2%) than for dysthymic disorders (2.1%) in children; while dysthymic disorders (6.9%) were more frequent than major depressive disorders (5.8%) in adolescents. These prevalence rates are consistent with those found in previous reviews and meta-analyses conducted among samples of TD youth (Birmaher et al., 1996; Merikangas, Nakamura, & Kessler, 2009; Polanczyk et al., 2015).

4.2 Moderators

The second objective of the current meta-analysis was to examine the source of heterogeneity in the reported prevalence of anxiety and depressive disorders. Due to the low number of studies including children or adolescents, the moderation analyses could not be conducted separately as a function of age.

Anxiety Disorders. Our findings suggest that prevalence of combined subtypes of anxiety disorders and obsessive-compulsive disorders are higher when the diagnosis is obtained by an interview rather than by multiple assessments. This result may be explained by the fact that a single method of data collection may be not sufficiently specific to screen for these types of psychological disorders among youth with ID. The complexity of the diagnosis of anxiety disorders among this population, which is partly related to their limited abilities to communicate internal states, has led to the recommendation

to rely on a multiple or multidimensional assessment (Pruijssers, van Meijel, & van Achterberg, 2011; Sullivan et al., 2006, 2011). Alternatively, it may also be hypothesized that an interview schedule is a more sensitive, and thus more appropriate, tool to identify psychological disorders among youth with ID where other means may be unable to do so.

In addition, higher prevalence of combined subtypes of anxiety disorders and obsessive-compulsive disorders (except for generalized anxiety disorder) were observed when the diagnosis was made using the DSM-IV or DSM-IV-TR criteria compared to the DSM-II, DSM-III, DSM-III-R, or the ICD-10 criteria. The results also showed that higher prevalence of combined subtypes of anxiety disorders were observed when parents were the only source of information rather than when both parents and youth were. This result may be explained by the fact that the parents' own distress may affect their report of their child's anxiety; or alternatively by the possibility that current diagnostic tools may be unable to correctly identify high levels of psychological disorders when reported by youth with ID themselves. The inclusion of youth as an additional source of information may certainly help the clinician to refine his/her decision. However, the process of obtaining these insights must be shown to be reliable for this population. Finally, no significant variations were found as a function of ID level, and geographic location.

Depressive Disorders. The findings showed that youth with borderline ID were significantly more at risk of having a depressive disorder than those with a moderate, severe, or unspecified ID. This may be due to the fact that the identification of depressive symptoms among youth with lower levels of intellectual functioning represents a challenging issue as a result of their limited abilities to communicate about their internal states and the inappropriateness of standard diagnostic criteria (Bailey & Andrews, 2003; Barnhill, 2008; Morin et al., 2010; Ross & Oliver, 2003; Smiley & Cooper, 2003). Therefore, some symptoms may not be recognized or identified correctly among youth with ID (Morin et al., 2010). To address this issue, the Royal College of Psychiatrists of the United Kingdom has decided to develop

adapted diagnostic criteria for use with adults with learning disabilities/mental retardation (DC-LD; Royal College of Psychiatrists, 2001). Additionally, other scholars (for reviews see: Janowsky & Davis, 2005; Morin et al., 2010) have also provided standard reference for the diagnosis of psychological disorders in persons with ID (Fletcher, Loschen, Stavrakaki, & First, 2007) or modified criteria for research (Clarke & Gomez, 1999). However, none of the studies included in the present review have used these criteria among youth with ID.

The findings of the current meta-analysis suggest that there is no substantial difference in the prevalence of anxiety and depressive disorders for youth with ID when compared to TD youth. This finding, however, may be attributable to the low quality of the studies reviewed. Methodological deficits in diagnoses impact on research that seeks to depict prevalence rates and thus inform practice. It is hypothesized that diagnostic overshadowing (Masi, 1998; Reiss, 2000) occurs whereby symptoms of psychological disorders do not result in a diagnosis but rather are attributed to the diagnosis of ID. Additionally, it is proposed that current diagnostic procedures may be ill-equipped to identify psychological disorders in youth with ID as their symptoms may not align with current diagnostic criteria for TD youth (Cooper, Melville, & Einfeld, 2003; Whitaker & Read, 2006).

4.3 Limitations and Directions for Future Studies

The present findings of this meta-analysis should also be interpreted with caution given several limitations. First, most of studies were conducted in North America or in Europe. Therefore, the magnitude of anxiety and depressive disorders in the Western Pacific region, African region, South-East Asia region, and Eastern Mediterranean region is underreported. Additionally, only a minority of studies have examined the prevalence according to sex, age, ID level, and additional diagnoses (autism spectrum disorder or genetic syndromes such as Down syndrome, Fragile X, etc.). Unlike the current meta-analysis, the majority of previous systematic reviews have included not only youth with ID but also youth with

additional diagnoses as a single combined group for purposes of analyses. Unlike the current meta-analysis, these reviews have typically found a higher prevalence of psychological disorders for this group compared to TD youth (e.g., Einfeld et al., 2011; Oeseburg et al., 2011). The present results suggest that these higher prevalence rates may in fact be due to these comorbid diagnoses, rather than to ID itself. Further studies, which take these variables into account, need to be undertaken in order to disentangle the relationship between ID, other associated diagnoses, and psychological disorders.

Second, only four studies (Baker et al., 2010; Emerson & Hatton, 2007; Green et al., 2015; Hardan & Sahl, 1997) compared the risk of anxiety/depressive disorders between youth with ID and TD youth. Therefore, it is unknown whether youth with ID were at greater risk of being diagnosed as having anxiety or depressive disorders than TD youth. Clearly, this is an important issue to consider in future studies.

Third, the time frame used in the diagnosis of anxiety and depressive disorders (i.e., current or lifetime) was reported in very few studies. Therefore, the role of this critically important factor in the prevalence estimates of anxiety or depressive disorders is unclear and should be more thoroughly investigated in future studies.

Fourth, only a few of the reviewed studies have examined specific subtypes of anxiety or depressive disorders among youth with ID. Additionally, none of the reviewed studies used diagnostic criteria adapted to individuals with ID. In future investigations, it would be interesting to determine the prevalence of anxiety or depressive disorders when using ID adapted diagnostic criteria compared to classic DSM or ICD criteria.

Finally, only five of the reviewed studies were population-based (Dekker & Koot, 2003; Emerson & Hatton, 2007; Gillberg et al., 1986; Jacobson, 1990; Rojahn et al., 1993). Therefore, serious concerns could be expressed regarding the representativeness of most of the samples, and by extension the value

of the reported prevalence estimates.

5. Conclusion

The present meta-analysis reveals that prevalence of anxiety and depressive disorders among youth with ID are generally similar to those found in TD children and adolescents. Additionally, findings reveal that diagnostic method (diagnostic criteria, types of informant) and youth's ID level may affect the likelihood of being diagnosed with an anxiety or depressive disorder. Significantly, the results of this meta-analysis highlight the potential inadequacy of current diagnostic criteria and diagnostic procedures to sufficiently recognize anxiety and depressive disorders in youth with ID. The comparison of psychological risk between youth with ID and TD youth still remains an understudied research area, and one hampered by diagnostic challenges with this population. Similarly, greater efforts are needed to ensure that clinicians optimize their capacity to appropriately diagnose comorbid anxiety and depression in youth with ID, and international psychological policy and practice need to be developed to meet this challenge. In sum, findings from this systematic review and meta-analysis will help to increase awareness among clinicians, service agencies, and policy makers about the nature of psychological disorders experienced by this vulnerable population. More importantly, these results should help key stakeholders to tailor resources to support youth with ID in the management of their anxiety and depressive disorders, and to better address fundamental issues of diagnosis that underpin and direct service provision and research internationally.

Author statement

Acknowledgements:

The preparation of this meta-analysis was supported by grants from the Social Sciences and Humanities Research Council of Canada (SSHRC; 430-2012-0091, 435-2014-0909) awarded to the first, third, fourth and sixth authors, and the Australian Research Council (ARC; DP140101559) awarded to the first, third and sixth authors.

Role of Funding Sources:

SSHRC and ARC had no role in the study design, collection, analysis or interpretation of the data, writing the manuscript, or the decision to submit the paper for publication.

Contributors:

All authors contributed to the design of the study. CM, SC, and GM operationalized the study, managed the literature search, selected the relevant studies, and extracted the information from the reviewed studies. CM and GM conducted the statistical analysis and assessed the quality of the reviewed studies. CM wrote the first draft of the manuscript. SC, DT, SB, GL, AJSM, GM critically reviewed and revised the manuscript. All authors contributed to and have approved the final manuscript.

Conflict of interest:

All authors declare that they have no conflicts of interest.

References

- Bailey, N. M., Andrews, T. M., 2003. Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation (DC-LD) and the diagnosis of anxiety disorders: A review. *J. Intellect. Disabil. Res.* 47(s1), 50-61.
- Baker, B. L., Neece, C. L., Fenning, R. M., Crnic, K. A., Blacher, J., 2010. Mental disorders in five-year-old children with or without developmental delay: Focus on ADHD. *J. Clin. Child. Psychol.* 39, 492–505.
- Barnhill, L. J., 2008. The diagnosis and treatment of individuals with mental illness and developmental disabilities: An overview. *Psychiatr. Q.* 79(3), 157-170.
- Baxter, A. J., Scott, K. M., Vos, T., Whiteford, H. A., 2013. Global prevalence of anxiety disorders: A systematic review and meta-regression. *Psychol. Med.* 43(5), 897-910.
- Begg, C. B., Mazumdar, M., 1994. Operating characteristics of a rank correlation test for publication bias. *Biometrics.* 50, 1088-1101.
- Birmaher, B., Ryan, N. D., Williamson, D. E., Brent, D. A., Kaufman, J., Dahl, R. E., et al., 1996. Childhood and adolescent depression: A review of the past 10 years. Part I. *J. Am. Acad. Child. Adolesc. Psychiatry.* 35(11), 1427-1439.
- Borenstein, M., Hedges, L., Higgins, J., Rothstein, H., 2005. *Comprehensive meta-analysis version 2*. Biostat, Englewood.
- Borthwick-Duffy, S. A., 1994. Epidemiology and prevalence of psychopathology in people with mental retardation. *J. Consult. Clin. Psychol.* 62, 17-27.
- Bradley, E. A., Ames, C. S., Bolton, P. F., 2011. Psychiatric conditions and behavioural problems in adolescents with intellectual disabilities: Correlates with autism. *Can. J. Psychiatry.* 56(2), 102-109.

- Buckles, J., Luckasson, R., Keefe, E., 2013. A systematic review of the prevalence of psychiatric disorders in adults with intellectual disability, 2003–2010. *J. Ment. Health. Res. Intellect. Disabil.* 6, 181-207.
- Chadwick, O., Kusel, Y., Cuddy, M., Taylor, E., 2005. Psychiatric diagnoses and behaviour problems from childhood to early adolescence in young people with severe intellectual disabilities. *Psychol. Med.* 35(5), 751-760.
- Clarke, D. J., Gomez, G. A., 1999. Utility of modified DCR-10 criteria in the diagnosis of depression associated with intellectual disability. *J. Intellect. Disabil. Res.* 43(5), 413-420.
- Cochran, W. G., 1950. The comparison of percentages in matched samples. *Biometrika.* 37, 256-266.
- Cooper, S. A., Melville, C. A., Einfeld, S. L., 2003. Psychiatric diagnosis, intellectual disabilities and diagnostic criteria for psychiatric disorders for use with adults with Learning Disabilities/Mental Retardation (DC-LD). *J. Intellect. Disabil. Res.* 47(Suppl. 1), 3–15.
- Costello, E. J., Egger, H. L., Copeland, W., Erkanli, A., Angold, A., 2011. The developmental epidemiology of anxiety disorders: Phenomenology, prevalence, and comorbidity, in: Silverman W. K., Field A. P. (Eds.). *Anxiety disorders in children and adolescents*, second ed. Cambridge University Press, Cambridge, pp. 56-75.
- Dekker, M. C., Koot, H. M., 2003. DSM-IV disorders in children with borderline to moderate intellectual disability. I. Prevalence and impact. *J. Am. Acad. Child. Adolesc. Psychiatry.* 42(8), 915-922.
- Duval, S., Tweedie, R., 2000. Trim and fill: A simple funnel-plot-based method of testing and adjusting for publication bias in meta-analysis. *Biometrics.* 56, 455-463.
- Egger, M., Smith, G. D., Schneider, M., Minder, C., 1997. Bias in meta-analysis detected by a simple, graphical test. *BMJ* 315, 629-634.

- Einfeld, S. L., Ellis, L. A., Emerson, E., 2011. Comorbidity of intellectual disability and mental disorder in children and adolescents: A systematic review. *J. Intellect. Dev. Disabil.* 36,137-143.
- Emerson, E., Hatton, C., 2007. Mental health of children and adolescents with intellectual disabilities in Britain. *BJPsych.* 191(6), 493-499.
- Fletcher, R., Loschen, E., Stavrakaki, C., First, M., 2007. DM-ID: Diagnostic manual-intellectual disability: a textbook of diagnosis of mental disorders in persons with intellectual disability. National Association for the Dually Diagnosed, Kingstom.
- Gillberg, C., Persson, E., Grufman, M., Themnér, U., 1986. Psychiatric disorders in mildly and severely mentally retarded urban children and adolescents: Epidemiological aspects. *BJPsych.* 149(1), 68-74.
- Gothelf, D., Goral, O., Avni, S., Stawski, M., Hartmann, I., Basel-Vanagaite, L., et al., 2008. Psychiatric morbidity with focus on obsessive-compulsive disorder in an Israeli cohort of adolescents with mild to moderate mental retardation. *J. Neural. Transm.* 115(6), 929-936.
- Green, S. A., Berkovits, L. D., Baker, B. L., 2015. Symptoms and development of anxiety in children with or without intellectual disability. *J. Clin. Child. Adolesc. Psychol.* 44(1), 137-144.
- Hardan, A., Sahl, R., 1997. Psychopathology in children and adolescents with developmental disorders. *Res. Dev. Disabil.* 18(5), 369-382.
- Hassiotis, A., Turk, J., 2012. Mental health needs in adolescents with intellectual disabilities: Cross-sectional survey of a service sample. *J. Appl. Res. Intellect. Disabil.* 25(3), 252-261.
- Hemmings, C., Deb, S., Chaplin, E., Hardy, S., Mukherjee, R., 2013. Review of research for people with ID and mental health problems: A view from the United Kingdom. *J. Ment. Health. Res. Intellect. Disabil.* 6, 127-158.

- Higgins, J. P., Thompson, S. G., Deeks, J. J., Altman, D. G., 2003. Measuring inconsistency in meta-analyses. *BMJ*. 327, 557.
- Hudson, C., Chan, J. B., 2002. Individuals with intellectual disability and mental illness: A literature review. *Aust J Soc Issues* 37, 31-49.
- Jacobson, J. W., 1990. Do some mental disorders occur less frequently among persons with mental retardation? *Am. J. Ment. Retard.* 94(6), 596-602.
- Janowsky, D. S., Davis, J. M., 2005. Diagnosis and treatment of depression in patients with mental retardation. *Curr. Psychiatry. Rep.* 7(6), 421-428.
- Khess, C. R., Dutta, I., Chakrabarty, I., Bhattacharya, P., Das, J., Kothari, S., 1998. Comorbidity in children with mental retardation. *Indian. J. Psychiatry.* 40(3), 289-294.
- Koskentausta, T., Iivanainen, M., Almqvist, F., 2002. Psychiatric disorders in children with intellectual disability. *Nord. J. Psychiatry.* 56(2), 126-131.
- Lakhan, R., 2013. The coexistence of psychiatric disorders and intellectual disability in children aged 3–18 years in the Barwani district, India. *ISRN Psychiatry.* 2013.
- Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P. A., et al., 2009. The PRISMA statement for reporting systematic reviews and meta-analysis of studies that evaluate health care interventions: explanation and elaboration. *PLoS Med.* 6, 1–6.
- Manor-Binyamini, I., 2010. The prevalence and characteristics of psychiatric disorders among adolescent Bedouin with mild to moderate intellectual disability. *Int. J. Spec. Educ.* 25(2), 26-33.
- Masi, G., 1998. Psychiatric illness in mentally retarded adolescents: Clinical features. *Adolesc.* 33(130), 425-433.

- Merikangas, K. R., Nakamura, E. F., Kessler, R. C., 2009. Epidemiology of mental disorders in children and adolescents. *Dialogues Clin. Neurosci.* 11(1), 7-20.
- Morin, D., Cobigo, V., Rivard, M., Lépine, M., 2010. Intellectual disabilities and depression: How to adapt psychological assessment and intervention. *Can. Psychol.* 51(3), 185-193.
- Myers, B. A., 1987. Psychiatric problems in adolescents with developmental disabilities. *J. Am. Acad. Child. Adolesc. Psychiatry.* 26(1), 74-79.
- Neyeloff, J. L., Fuchs, S. C., Moreira, L. B., 2012. Meta-analyses and forest plots using a Microsoft Excel spreadsheet: Step-by-step guide focusing on descriptive data analysis. *BMC. Res. Notes.* 5, 52.
- Oeseburg, B., Dijkstra, G. J., Groothoff, J. W., Reijneveld, S. A., Jansen, D. E. C., 2011. Prevalence of chronic health conditions in children with intellectual disability: A systematic literature review. *Intellect. Dev. Disabil.* 49, 59-85.
- Philips, I., Williams, N., 1975. Psychopathology of mental retardation: A study of 100 mentally retarded children: I. Psychopathology. *Am. J. Psychiatr.* 132(12), 1265-1271.
- Polanczyk, G. V., Salum, G. A., Sugaya, L. S., Caye, A., Rohde, L. A., 2015. Annual Research Review: A meta-analysis of the worldwide prevalence of mental disorders in children and adolescents. *J. Child. Psychol. Psychiatry.* 56(3), 345-365.
- Pruijssers, A., van Meijel, B., van Achterberg, T., 2011. A case report for diagnosing anxiety in people with intellectual disability: The role of nurses in the application of a multidimensional diagnostic guideline. *Perspect. Psychiatr. Care.* 47(4), 204-212.
- Reardon, T. C., Gray, K. M., Melvin, G. A., 2015. Anxiety disorders in children and adolescents with intellectual disability: Prevalence and assessment. *Res. Dev. Disabil.* 36, 175-190.

- Reinherz, H. Z., Paradis, A. D., Giaconia, R. M., Stashwick, C. K., Fitzmaurice, G., 2003. Childhood and adolescent predictors of major depression in the transition to adulthood. *Am. J. Psychiatry.* 160(12), 2141-2147.
- Reiss, S., 2000. A mindful approach to mental retardation. *J. Soc. Issues.* 56, 65–80.
- Reiss, S., 1982. Psychopathology and mental retardation: Survey of a developmental disabilities mental health program. *Ment. Retard.* 20(3), 128-132.
- Rojahn, J., Borthwick-Duffy, S. A., Jacobson, J. W., 1993. The association between psychiatric diagnoses and severe behavior problems in mental retardation. *Ann. Clin. Psychiatry.* 5(3), 163-170.
- Ross, E., Oliver, C., 2003. The assessment of mood in adults who have severe or profound mental retardation. *Clin. Psychol. Rev.* 23(2), 225-245.
- Royal College of Psychiatrists, 2001. DC-LD [Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation]. Gaskell Pres, London.
- Royston, R., Howlin, P., Waite, J., Oliver, C., 2017. Anxiety disorders in Williams syndrome contrasted with intellectual disability and the general population: A systematic review and meta-analysis. *J. Autism. Dev. Disord.* 47(12), 3765-3777.
- Roza, S. J., Hofstra, M. B., van der Ende, J., Verhulst, F. C., 2003. Stable prediction of mood and anxiety disorders based on behavioral and emotional problems in childhood: A 14-year follow-up during childhood, adolescence, and young adulthood. *Am. J. Psychiatry.* 160(12), 2116-2121.
- Shamliyan, T., Ansari, M. T., Raman, G., Berkman, N., Grant, M., Janes, G., et al., 2013. Development and implementation of the standards for evaluating and reporting epidemiologic studies on chronic disease incidence or prevalence. *Am. J. Public. Health. Res.* 1(7), 183-190.

- Smiley, E., Cooper, S. A., 2003. Intellectual disabilities, depressive episode, diagnostic criteria and Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation (DC-LD). *J. Intellect. Disabil. Res.* 47(s1), 62-71.
- Strømme, P., Diseth, T. H., 2000. Prevalence of psychiatric diagnoses in children with mental retardation: data from a population-based study. *Dev. Med. Child. Neurol.* 42(4), 266-270.
- Stroup, D. F., Berlin, J. A., Morton, S. C., Olkin, I., Williamson, G. D., Rennie, D., et al., 2000. Meta-analysis of observational studies in epidemiology: A proposal for reporting. *JAMA.* 283, 2008-2012.
- Sullivan, W. F., Berg, J. M., Bradley, E., Cheetham, T., Denton, R., Heng, J., et al., 2011. Primary care of adults with developmental disabilities. *Can. Fam. Physician.* 57(5), 541-553.
- Sullivan, W. F., Heng, J., Cameron, D., Lunsky, Y., Cheetham, T., Hennen, B., et al., 2006. Consensus guidelines for primary health care of adults with developmental disabilities. *Can. Fam. Physician.* 52(11), 1410-1418.
- Temtek, S., Uneri, O. S., Goker, Z., Dinc, G., Hekim, O., 2015. Psychotropic medication use in children and adolescents with mental retardation (MR). *Am. J. Psychiatry. Neurosci.* 3(6), 111-115.
- van Steensel, F. J., Bögels, S. M., Perrin, S. 2011. Anxiety disorders in children and adolescents with autistic spectrum disorders: a meta-analysis. *Clin. Child. Fam. Psychol. Rev.* 14(3), 302-317.
- Whitaker, S., Read, S., 2006. The prevalence of psychiatric disorders among people with intellectual disabilities: an analysis of the literature. *J. Appl. Res. Intellect. Disabil.* 19, 330-345.
- Yoo, J. H., Valdovinos, M. G., Schroeder, S. R., 2012. The epidemiology of psychopathology in people with intellectual disability: A forty-year review. *Int. Rev. Res. Dev. Disabil.* 42, 31-56.

Endnote

¹ Other reviews (e.g., Borthwick-Duffy, 1994; Buckles, Luckasson, & Keefe, 2013; Hemmings, Deb, Chaplin, Hardy, & Mukherjee, 2013; Yoo, Valdovinos, & Schroeder, 2012) focused on the prevalence of psychopathology, psychiatric disorders, or mental health disorders among people with ID, but they did not synthesize their results separately for children-adolescents versus adults.

² In studies focusing specifically on participants with ID, those including participants with borderline ID were considered because these participants also face “*substantially elevated cognitive and morbidity risks as well as problems in adaptive behavior*” (Oeseburg et al., 2011, p. 60).

³ This category includes studies using a general (or unspecified) measure of anxiety disorder or providing an overall prevalence rate for combined subtypes of anxiety disorders.

⁴ This category includes studies using a general (or unspecified) measure of depressive disorder or providing an overall prevalence rate for combined subtypes of depressive disorders.

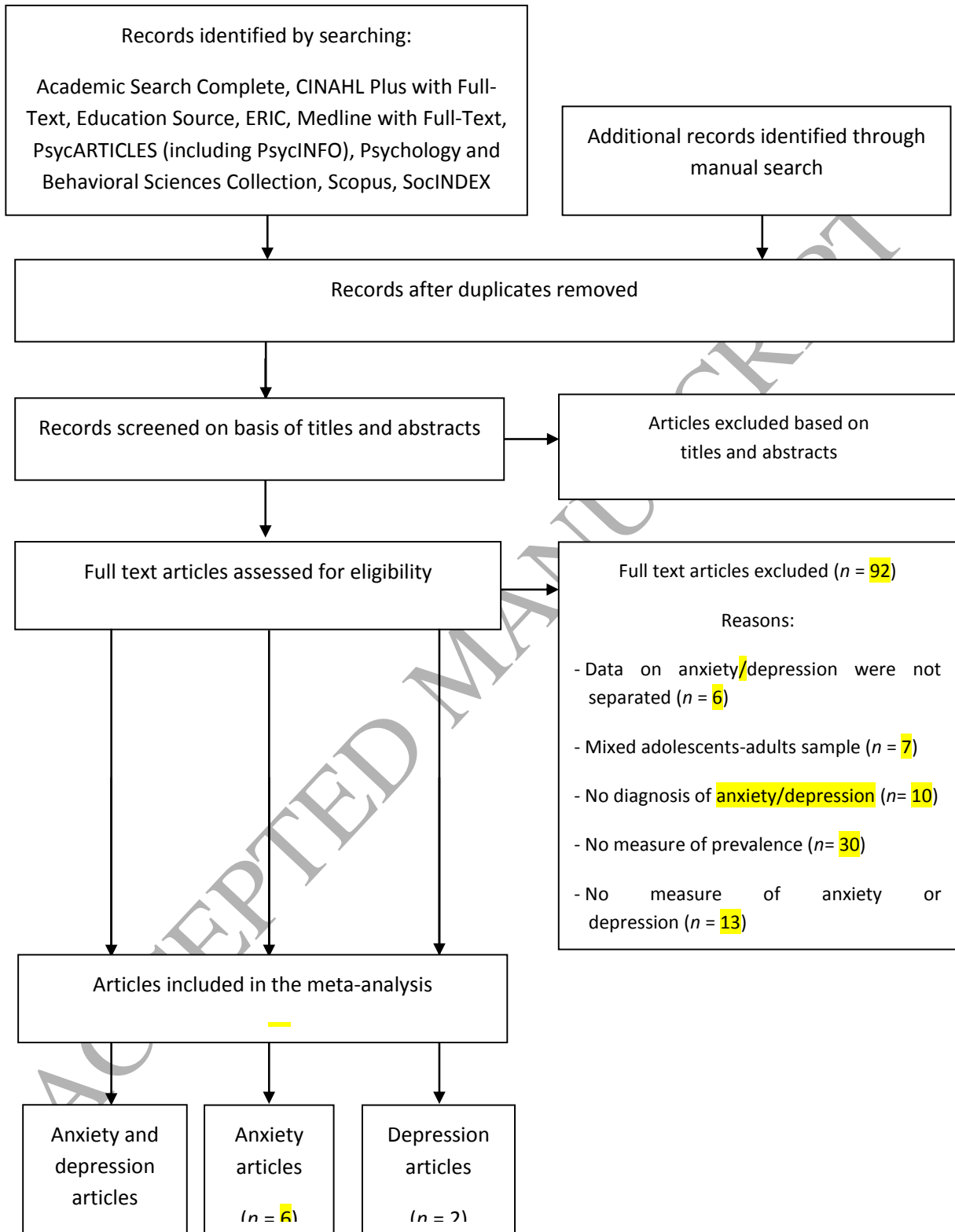


Figure 1. Results of search based on the PRISMA statement (Liberati et al., 2009)

Note. ID = intellectual disability.

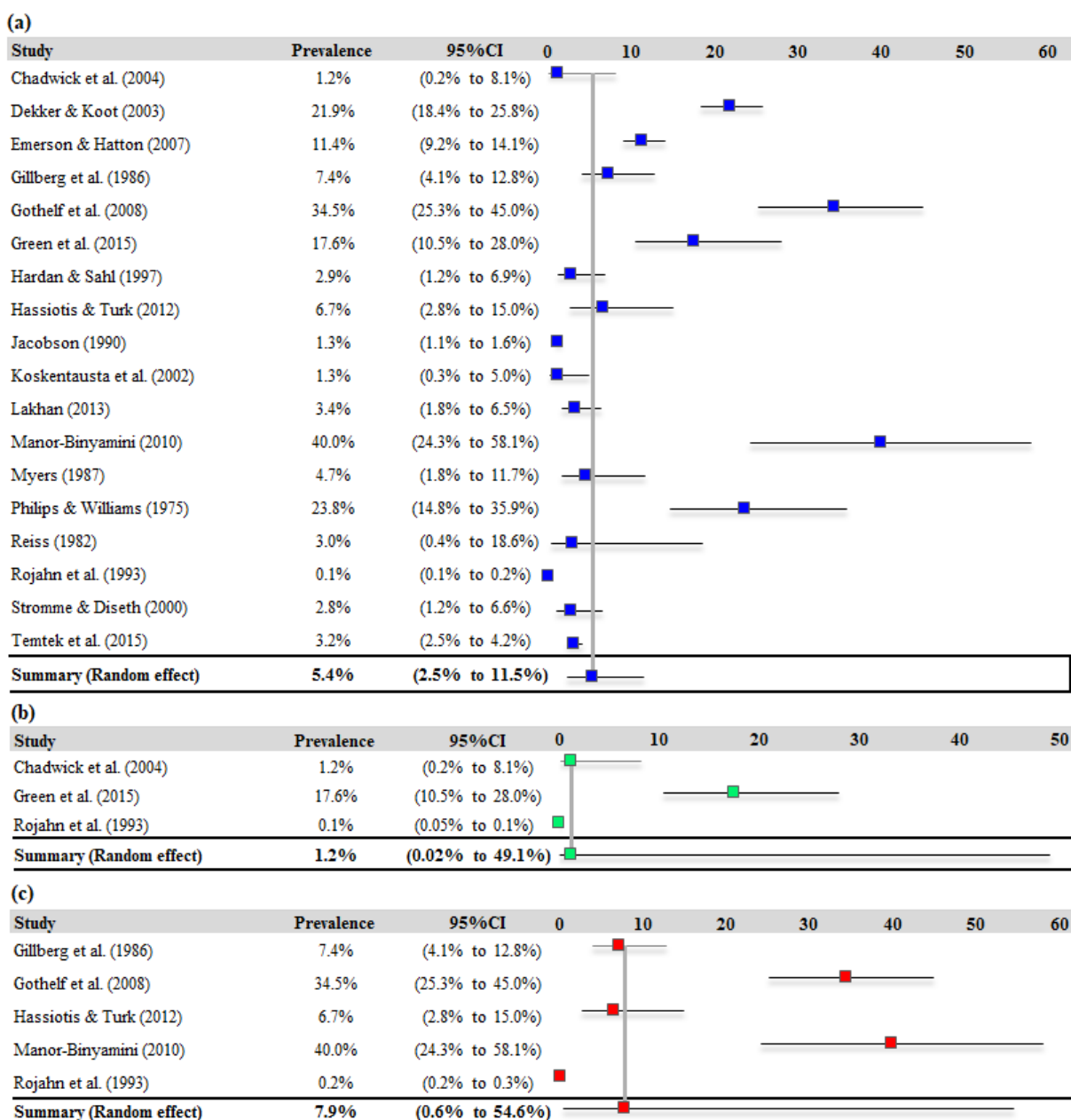


Figure 2. Forest plot of random-effects pooled prevalence estimates of combined subtypes of anxiety disorders in (a) all reviewed samples (mixed, children, and adolescents), (b) children samples, and (c) adolescent samples

Note. For the estimation of the overall sample, the prevalence estimates of the children and the adolescent samples from Rojahn et al.'s (1993) study have been combined.

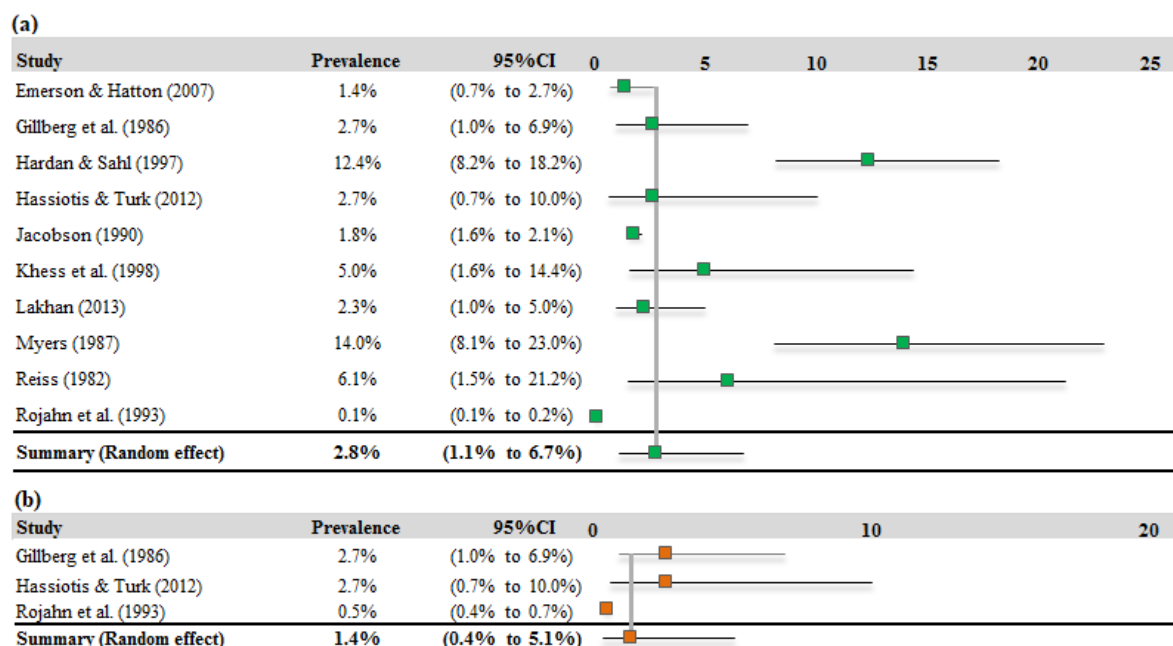


Figure 3. Forest plot of random-effects pooled prevalence estimates of combined subtypes of depressive disorders in (a) all reviewed samples (mixed, children, and adolescents), and (b) adolescent samples

Note. For the estimation of the overall sample, the prevalence estimates of the children and the adolescent samples from Rojahn et al.'s (1993) study have been combined

Anxiety, Depression, and Intellectual Disabilities

38

Study	Country	Design	Recruitment setting	Type of samples	ID sample	TD sample
-------	---------	--------	---------------------	-----------------	-----------	-----------

Table 1*Main Characteristics of the Studies Included in the Meta-Analysis*

					Sample size (N)	% of boys	Age range	ID level	Yes/No	Sample size (N)
Baker et al. (2010)	USA	Cohort	Service agencies	CHILD	95 ^a	60	5	NM	Yes	141
Bradley et al. (2011)	Canada	CS	NM	ADOS	72 ^b	NM	14-20	NM	No	
Chadwick et al. (2005)	UK	Cohort	Special school	CHILD	82 ^c	56	4-11	Severe	No	
Dekker & Koot (2003)	Netherlands	Cohort	Special school	Mixed	474	62	7-20	Mild-Moderate	No	
Emerson & Hatton (2007)	UK	CC	National survey - ONS (1999, 2004)	Mixed	641	66	5-15	NM	Yes	17,774
Gillberg et al. (1986)	Sweden	CS	Service agencies, schools	ADOS	149	62	13-17	Mild-Severe	No	
Gothelf et al. (2008)	Israel	CS	Special school	ADOS	87 ^d	53	12-21	Mild-Moderate	No	
Green et al. (2015)	USA	Cohort	Service agencies	CHILD	74 ^a	61	5	Borderline-Moderate	Yes	116
Hardan & Sahl (1997)	USA	CC	Service agencies, community, school	Mixed	170 ^e	NM	3-19	Borderline-Profound	Yes	63
Hassiotis & Turk (2012)	UK	CS	Service agencies	ADOS	75	64	12-19	Mild-Profound	No	
Jacobson (1990)	USA	CS	Information system	Mixed	9,876	59	0-21	Mild-Profound	No	
Khess et al. (1998)	India	CS	Child psychiatric unit	Mixed	60	68	NM	Mild-Profound	No	
Koskentausta et al. (2002)	Finland	CS	Rehabilitation center, hospitals, special schools	Mixed	155	59	6-13	Mild-Profound	No	
Lakhan (2013)	India	CS	Service agencies	Mixed	262 ^f	NM	3-18	Borderline-Profound	No	
Manor-Binyamini (2010)	Israel	CS	Special school	ADOS	30	70	12-21	Mild-Moderate	No	
Myers (1987)	USA	CS	Hospital	Mixed	86 ^g	53	10-21	Mild-Profound	No	

Anxiety, Depression, and Intellectual Disabilities

40

Philipps & Williams (1975)	USA	CS	Psychiatric clinic	Mixed	62 ^h	61	0-18	Borderline-Severe	No
Reiss (1982)	USA	CS	Service agencies	Mixed	33	70	6-20	Mild-Profound	No
Rojahn et al. (1993)	USA	CS	Service agencies	CHILD, ADOS	43,738 ⁱ	NM	0-20	Mild-Profound	No
Stromme & Diseth (2000)	Norway	CS	Referred to the study	Mixed	178	58	8-13	Mild-Severe	No
Temtek et al. (2015)	Turkey	CS	Hospital	Mixed	1,572	60	6-18	Mild-Profound	No

Note. ADOS = adolescent sample; CC = case control; CHILD = children sample; CS = cross-sectional; N = number; NM = not mentioned; TD = typically developing; UK = United Kingdom; USA = United States of America. ^aThe data are those from age 5 only, and the participants with a borderline intellectual disability were included. ^bThis sample includes youth with intellectual disabilities and youth with intellectual disabilities and autism; ^cThe data are those from the initial study. ^dOnly the current prevalence's data have been used. ^eThe participants with a borderline and an unspecified intellectual disability were included in the sample. ^fThe participants with a borderline intellectual disability were included in the sample. ^gThe participants with a borderline intellectual disability were not included in the sample because they were reported together with those without an intellectual disability. ^hThis sample only comprised children with intellectual disabilities without psychiatric disorders. ⁱThe data from the New York State sample were not included because of the probable overlap with Jacobson's (1990) sample.

Table 2*Diagnostic Information and Types of Anxiety and Depressive Disorders of the Studies Included in the Meta-Analysis*

Study	Diagnostic information			Types of anxiety disorders	Types of depressive disorders
	Informant	Method	Criteria		
Baker et al. (2010)	P	Interview-S (DISC-IV)	DSM-IV-TR	Not included ^a	MDD, DD
Bradley et al. (2011)	P	Interview-S (SAPPA)	ICD-10	GAD	Not examined
Chadwick et al. (2005)	P	Medical records	NM	Combined ^a	Not examined
Dekker & Koot (2003)	C, P	Interview-S (DISC-IV)	DSM-IV	Combined ^a , AGO, GAD, OCD, PDWA, PDWIA, PTSD, SAD, SP, SPHOBIA	MDD, DD
Emerson & Hatton (2007)	C, Y, T	Multiple assessments	ICD-10	Combined ^a , AGO, GAD, OCD, PD, PTSD, SAD, SP, SPHOBIA	Combined ^b
Gillberg et al. (1986)	P, Y	Multiple assessments	DSM-III	Combined ^c	Combined ^d
Gothelf et al. (2008)	P	Interview-S (K-SADS-PL)	DSM-IV-TR	Combined ^a , GAD, OCD, PD, PTSD, SAD, SP, SPHOBIA	MDD, DD
Green et al. (2015)	P	Interview-S (DISC-IV)	DSM-IV	Combined ^a , GAD, SAD, SP	Not examined
Hardan & Sahl (1997)	NM	Medical records	DSM-III-R	Combined ^e , OCD, PTSD	Combined ^f
Hassiotis & Turk (2012)	Y	Interview-SM	ICD-10	Combined ^a , SP	Combined ^b
Jacobson (1990)	NM	Medical records	DSM-II	Combined ^g	Combined ^h
Khess et al. (1998)	NM	Multiple assessments	ICD-10	Not examined	Combined ⁱ
Koskentausta et al. (2002)	NM	Medical records	ICD-10	Combined ^j	MDD
Lakhan (2013)	P, Y	Multiple assessments	ICD-10	Combined ^k , OCD	Combined ^l

Manor-Binyamini (2010)	P	Interview-S (K-SADS-PL)	DSM-IV-TR	Combined ^a , OCD, SPHOBIA	Not examined
Myers (1987)	P	Multiple assessments	DSM-III	Combined ^m	Combined ⁿ
Philipps & Williams (1975)	NM	Medical records	DSM-II	Combined ^o	Not examined
Reiss (1982)	C, P, Y	Multiple assessments	NM	Combined ^p	Combined ⁱ
Rojahn et al. (1993)	NM	Medical records	DSM-III-R	Combined ^a	Combined ^q
Stromme & Diseth (2000)	P, Y	Multiple assessments	ICD-10	Combined ^f	Not examined
Temtek et al. (2015)	NM	Medical records	DSM-IV-TR	Combined ^a	Not examined

Note. AGO = agoraphobia without panic disorder; C = caregiver; DD = dysthymic disorder; DISC-IV = Diagnostic Interview Schedule for Children – 4th version; DSM-II = diagnostic and statistical manual of mental disorders – 2nd edition; DSM-III = diagnostic and statistical manual of mental disorders – 3rd edition; DSM-III-R = diagnostic and statistical manual of mental disorders – revised 3rd edition; DSM-IV = diagnostic and statistical manual of mental disorders – 4th edition; DSM-IV-TR = diagnostic and statistical manual of mental disorders – text revision of the 4th edition; GAD = generalized anxiety disorder; ICD-10 = international statistical classification of diseases and related health problems – 10th revision; Interview-S = structured interview; Interview-SM = semi-structured interview; K-SADS-PL = Schedule for Affective Disorders and Schizophrenia for School-Aged Children, Present and Lifetime; MDD = major depressive disorder; NM = not mentioned; OCD = obsessive-compulsive disorder; P = parents; PD = panic disorder; PDWA = panic disorder without agoraphobia; PDWIA = panic disorder with agoraphobia; PTSD = posttraumatic stress disorder; SAD = separation anxiety disorder; SAPP = Schedule for the Assessment of Psychiatric Problems associated with Autism (and other developmental disorders); SP = social phobia; SPHOBIA = specific phobia; T = teacher; Y = youth. ^aThese data were not included because of the probable overlap with the Green et al.'s (2015) sample. [^]Given that the nature of phobia was not specified these data were not included. [^]anxiety disorder. [^]Depressive disorder. [^]Emotional disorder (anxiety and fear without loss of reality-sense were the most incapacitating symptoms). [^]Depressive syndrome. [^]Overanxious disorder, separation anxiety disorder, anxiety disorder, not otherwise specified, phobias, and panic disorder. [^]Depressive major, depressive disorder, recurrent, depressive disorder, not otherwise specified, and dysthymia. [^]Neurosis (anxiety and phobic disorders). [^]Nonpsychotic organic brain syndromes. [^]Depression. [^]Emotional disorders with onset specific to childhood. [^]Anxiety, obsessive compulsive disorder. [^]Depression. [^]Separation anxiety disorder, overanxious, obsessive compulsive disorder. [^]Depression, major affective disorder. [^]Neurotic traits (phobias, obsessive-compulsive symptoms, mannerisms and preoccupations, anxiety reactions). [^]Phobia, anxiety neurosis. [^]Affective disorders. [^]Anxiety, phobic, obsessive-compulsive.

Anxiety, Depression, and Intellectual Disabilities

	PDWIA	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	PTSD	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	SAD	1	17.6%	(10.5% to 21.8%)	-5.06	<.001	0	0	1	0	NA	NA	NA		
	SP	1	2.7%	(0.7% to 10.2%)	-5.00	<.001	0	0	1	0	NA	NA	NA		
	SPHOBIA	-	-	-	-	-	-	-	-	-	-	-	-	-	-
Adolescents	Combined	5	7.9%	(0.6% to 54.6%)	-1.82	.068	499.7	4	<.001	99	.50	.12	3 missing	0.5%	(0.03% to 7.8%)
	AGO	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	GAD	2	8.7%	(2.4% to 27.1%)	-3.39	<.001	4.4	1	.03	78	NA	NA	NA		
	OCD	2	12.0%	(7.2% to 19.2%)	-7.00	<.001	0.1	1	.79	0	NA	NA	NA		
	PD	1	0.6%	(0.04% to 8.4%)	-3.64	<.001	0	0	1	0	NA	NA	NA		
	PDWA	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	PDWIA	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	PTSD	1	3.4%	(1.1% to 10.2%)	-5.67	<.001	0	0	1	0	NA	NA	NA		
	SAD	1	5.7%	(2.4% to 13.1%)	-6.07	<.001	0	0	1	0	NA	NA	NA		
	SP	2	5.4%	(1.9% to 14.6%)	-5.11	<.001	2	2	.16	50	NA	NA	NA		
	SPHOBIA	2	19.8%	(11.8% to 31.3%)	-4.47	<.001	1.6	1	.21	38	NA	NA	NA		

Note. AGO = agoraphobia without panic disorder; B-M test = Begg and Mazumdar rank correlation test; DT-FT = Duval and Tweedie's trim and fill; Egger-T = Egger's test of the intercept; GAD = generalized anxiety disorder; OCD = obsessive-compulsive disorder; PD = panic disorder; PDWA = panic disorder without agoraphobia; PDWIA = panic disorder with agoraphobia; PTSD = posttraumatic stress disorder; SAD = separation anxiety disorder; SP = social phobia; SPHOBIA = specific phobia.

Table 4*Results of the Random Effects Models, Tests for heterogeneity, and Publication Bias Across Samples and Types of Depressive Disorders*

Samples	Types of depressive disorders	N studies	Random effects models				Tests for heterogeneity				Publication bias					
			Prevalence	95% CI		Z-value	p	Q	df	p	I ² (%)	B-M test	Egger-T	DT-TF	Prevalence	95% CI
All	Combined	10	2.8%	(1.1% to	6.7%)	-7.58	0.00	242.3	9	<.001	96	.24	.31	3 missing	1.6%	(0.6% to 3.7%)
	DD	3	3.4%	(1.5% to	7.4%)	-7.95	<.001	5.2	2	.07	62	.50	.45	No missing		
	MDD	4	2.5%	(1.2% to	5.4%)	-8.98	<.001	6.8	3	.08	56	.50	.41	No missing		
Children	Combined	1	0.03%	(0.02% to	0.06%)	-24.00	<.001	0	0	1	0	NA	NA	NA		
	DD	1	2.1%	(0.5% to	8.0%)	-5.37	<.001	0	0	1	0	NA	NA	NA		
	MDD	1	3.2%	(1.0% to	9.4%)	-5.85	<.001	0	0	1	0	NA	NA	NA		
Adolescents	Combined	3	1.4%	(0.4% to	5.1%)	-6.31	0.00	14.5	2	<.001	86	.50	.07	2 missing	0.5%	(0.2% to 1.6%)
	DD	1	6.9%	(3.1% to	14.5%)	-6.15	<.001	0	0	1	0	NA	NA	NA		
	MDD	1	5.7%	(2.4% to	13.1%)	-6.07	<.001	0	0	1	0	NA	NA	NA		

Note. B-M test = Begg and Mazumdar rank correlation test; DD = dysthymic disorder; DT-TF = Duval and Tweedie's trim and fill; Egger-T = Egger's test of the intercept; MDD = major depressive disorder.

Anxiety, Depression, and Intellectual Disabilities

47

Lakhan (2013)	○	○	●	○	○	●	●	○	○	○
Manor-Binyamini (2010)	○	○	○	○	○	●	●	○	○	○
Myers (1987)	○	○	○	○	○	●	○	○	○	○
Philipps & Williams (1975)	○	○	○	○	○	●	○	○	○	○
Reiss (1982)	○	○	○	○	○	●	○	○	○	○
Rojahn et al. (1993)	●	○	●	○	○	○	○	○	○	○
Stromme & Diseth (2000)	○	○	●	○	○	●	●	○	○	○
Temtek et al. (2015)	○	○	●	○	○	○	○	○	○	○
Total	6/21	4/21	9/21	1/21	7/21	15/21	11/21	5/21	4/21	2/21

Note. ● = met the criteria; ○ = did not meet the criteria; Total = number of studies meeting each quality criteria