# Anxiety and depression symptoms in children and adolescents who stutter: A systematic review and meta-analysis

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## Abstract

***Purpose***. To investigate whether there are elevated symptoms of anxiety or depression in children and adolescents (aged 2–18 years) who stutter, and to identify potential moderators of increased symptom severity.

***Method***. We conducted a pre-registered systematic review of databases and grey literature; 13 articles met criteria for inclusion. A meta-analysis using Robust Variance Estimation (RVE) was conducted with 11 cohort studies comparing symptoms of anxiety in children and adolescents who do and do not stutter. Twenty-six effect sizes from 11 studies contributed to the summary effect size for anxiety symptoms (851 participants). Meta-analysis of depression outcomes was not possible due to the small number of studies.

***Results***. The summary effect size indicates that children and adolescents who stutter present with increased anxiety symptoms ($g$ = 0.42) compared to non-stuttering peers. There were insufficient studies to robustly analyse depression symptoms and qualitative review is provided. No significant between-group differences were reported in any of the depression studies.

***Conclusions***. Preliminary evidence indicates elevated symptoms of anxiety in some children and adolescents who stutter relative to peers. There was a tendency towards higher depression scores in this population, though reported between-group differences did not reach statistical significance. These findings require replication in larger, preferably longitudinal studies that

consider factors that may moderate risk. Nevertheless, our findings highlight a need for careful monitoring of mental health and well-being in young people who stutter.

# Introduction

Stuttering is a neurodevelopmental condition characterised by disturbances in speech fluency, comprising core behaviours (repetitions, blocks and prolongations), in which secondary behaviours (physical concomitants and substitutions) and negative attitudes may be present (Guitar, 2014). It is estimated that between 5% and 8% of children will stutter at some point in development (Yairi & Ambrose, 2013), while prevalence of persistent stuttering is estimated at 1% (Bloodstein & Bernstein-Ratner, 2008). Stuttering can have broad impacts on quality of life, though outcomes are variable (Craig et al., 2009). Some adults who stutter report increased fear of negative evaluation, heightened communication apprehension, and poor self-perceptions of communication competence (Blood et al., 2001; Messenger et al., 2004). There is also evidence of poorer psychosocial outcomes, including anxiety, amongst adolescents and adults who stutter, particularly in those who experienced childhood bullying (Blood & Blood, 2016; Cooke & Howell, 2014). While there have been a number of studies documenting the association between stuttering and anxiety, and to a lesser extent stuttering and depression, in adults (Craig & Tran, 2014; Iverach et al., 2009a; Livingstone-Pountney & Mitrevski, 2019), much less is known about these relationships in children and adolescents. The aim of this study is to evaluate evidence concerning increased risk for heightened symptoms of anxiety and depression in children and adolescents who stutter.

## Constructs of Anxiety and Depression

Anxiety is a complex psychological construct that comprises cognitive-verbal, behavioural and physiological components and is characterised by negative emotion that occurs in response to perceived threat (Essau et al., 2013; Menzies et al., 1999; Smith et al., 2014). The cognitive components of anxiety include negative thoughts and beliefs about upcoming events; behavioural aspects include a desire to escape and avoidance of situations; while physiological components comprise activation of the sympathetic nervous system resulting in physical sensations such as muscle tension, sweating and heart palpitations (Essau et al., 2013; Smith et al., 2014).

Anxiety consists of both state and trait elements. State anxiety is described as a transitory state of arousal that an individual experiences when faced by a potentially demanding or dangerous situation; trait anxiety is considered a permanent personality characteristic reflecting individual differences in how people respond to potentially threatening situations (Endler & Parker, 1990). Consequently, state anxiety is context-specific and may be elicited by factors relating to social interaction, whereas trait anxiety occurs independently of situational factors (Diehl et al., 2019). However, the distinction between state and trait anxiety is not as clear as it might seem. It is argued that both state and trait anxiety are multidimensional, and that levels of state anxiety depend also on the person (or trait) and the context (Endler & Kocovski, 2001).

Anxiety disorders involve abnormal levels of anxiety and are diagnosed when symptoms become persistent, excessive, and daily functioning is negatively impacted, which may be observed in self-report, behavioural, cognitive and physiological responses and underlying neural functioning (APA 2013; Craske et al., 2009). Social anxiety disorder is characterised by fear or avoidance of social interactions and situations that may result in scrutiny, while generalised anxiety disorder is associated with persistent and excessive worry in multiple contexts (APA, 2013).

Similarly to anxiety, depression is characterised by behavioural, cognitive and physical symptoms (Huberty, 2012). In the general population, anxiety and depression often co-occur (Huberty, 2012, Lewinsohn et al., 1997). Depression is characterised by prolonged sad, empty or irritable mood, as well as somatic and cognitive changes that have a substantial functional impact (APA 2013). Peck (2013) argues depression is a “multifaceted phenomenon with a variety of psychological and motor aspects” (p.408). Social anxiety disorder during adolescence is one risk factor for later onset of depression (Stein et al., 2001).

## Anxiety and depression in stuttering

Earlier narrative reviews exploring the relationship between anxiety and stuttering concluded that evidence of an association was weak (Ingham, 1984; Menzies, Onslow & Packman, 1999). Menzies et al. (1999) cited five sources of bias that contributed to difficulty interpreting the role of anxiety in stuttering: (i) failure to take account of the multidimensional nature of anxiety; (ii) inclusion of small sample sizes that led to insufficient statistical power; (iii) treatment status of participants (i.e. previous treatment for stuttering may have reduced anxiety); (iv) the speaking tasks employed when measuring or manipulating anxiety; and (v) the measures used to assess trait anxiety in particular. Iverach et al. (2011) reviewed studies published since Menzies et al.’s (1999) original review, with specific focus on these five methodological issues. They concluded that more recent studies offered stronger evidence of a relationship between stuttering and anxiety, particularly social anxiety, although they noted that many methodological issues remained, especially in relation to study design, statistical power and use of appropriate assessment measures.

The findings of two meta-analyses indicate that persistent stuttering in adults is associated with significantly elevated trait anxiety (*g* = 0.57) and social anxiety (*g* = 0.82) relative to non-stuttering adults (Craig & Tran, 2014). Additionally, adults who stutter appear to be at increased risk of meeting diagnostic criteria for clinical anxiety disorders, particularly social anxiety disorder (Iverach et al., 2009a; Iverach & Rapee, 2014).

While studies have shown elevated depression symptoms in some adolescents and young adults who stutter (e.g. Briley, Gerlach & Jacobs, 2021; Doruk et al., 2008), others have not (e.g. Bray et al., 2008). Recently, Livingstone-Pountney and Mitrevski (2019) provided a narrative review of the existing literature reporting on depression symptoms in adults and adolescents (11–18 years) who stutter. The review highlighted inconsistency within the adult literature, with half the studies reporting a significant relationship between stuttering and depression and half reporting no significant relationship. None of the three adolescent studies reported increased symptoms of depression. The authors noted inconsistency in controlling for comorbid conditions, participant treatment status and stuttering severity ratings across included studies, which has implications for interpretation, generalisability and design of future studies (Livingstone-Pountney & Mitrevski, 2019). The current study extends this work in important ways by providing a systematic review of available evidence that covers a broader age range and mandates inclusion of a non-stuttering comparison group in order to determine the magnitude of difference in symptom scores.

## Anxiety and depression in children and adolescents

It is estimated that half of all lifetime cases of poor mental health have onset prior to 14 years of age, with three-quarters occurring by 24 years of age (Kessler et al., 2005). Among children and adolescents, the worldwide prevalence of any anxiety disorder is 6.5%, while depressive disorder is estimated at 3.4% (Polanczyk et al., 2015). There are concerns that mental health issues, particularly anxiety and depression, are increasing among adolescents in high-income countries (Bor et al., 2014; Collishaw, 2015; Patalay & Gage, 2019; Pitchforth et al., 2018). Given the documented co-morbidity between anxiety and depression in population studies of children and adolescents (Cole et al., 1998; Lewinsohn et al., 1997; Whalen et al., 2017), and research showing higher anxiety symptoms predict higher depression symptoms among adolescents who stutter (Iverach et al., 2017), symptoms of both anxiety and depression are of interest in the current review.

Evidence of elevated anxiety symptoms in children who stutter appears to be variable and narrative reviews have examined potential onset and additional risk factors associated with elevated symptom levels of anxiety (see Alm, 2014; Smith et al., 2014). These reviews indicate that children who stutter do not appear to be at increased familial risk of anxiety or have temperament traits that predispose to anxiety. However, children who stutter are more likely to be exposed to negative peer reactions, bullying and stereotyping, which may increase risk of anxiety (Smith et al., 2014). Both reviews found limited evidence of elevated anxiety symptoms or temperament traits in pre-school children who stutter, but suggested that symptoms of anxiety may increase in later childhood. Smith et al. (2014) suggested that ‘environmental’ risk factors may manifest during adolescence, which coincides with increased prevalence of anxiety and social anxiety disorder in the general non-stuttering population. This causal hypothesis posits that anxiety in adults who stutter is a consequence of increasing self-awareness and exposure to negative reactions from peers, particularly as academic, vocational, social and interpersonal demands increase during adolescence and early adulthood.

Symptoms of anxiety and depression are often elevated in children and adolescents with speech and language disorders such as developmental language disorder (Beitchman, et al., 2001; Yew & O'Kearney, 2013). Children who stutter have been reported to have a higher incidence of co-occurring language disorders (Briley & Ellis, 2018), though not all studies have identified greater rates of co-morbidity (Nippold, 2019). Following review of the existing literature, Alm (2014) concluded that there may be elevated risk for social anxiety amongst children and adolescents who stutter with co-occurring deficits. Elevated risk of anxiety may also be a consequence of living with stuttering, as fear of stuttering could be perceived as demanding and frustrating in social situations. The higher level of anxiety in adults who stutter (Craig & Tran, 2014), and the potential for increased exposures to known risk factors (bullying, broader communication challenges) makes it relevant to ask if children and adolescents who stutter also have elevated anxiety and/or depression symptoms compared to children and adolescents who do not stutter. If there were elevated symptoms of anxiety and/or depression among children who stutter, it would be useful to determine whether age, stuttering severity and co-morbidity mitigate symptom severity, as this could be important for clinical services.

## The current review

This systematic review evaluates associations between stuttering and symptoms of anxiety and depression in people aged under 18 years. The review asks:

• Are children and adolescents who stutter more likely to present with elevated symptoms of anxiety or depression relative to peers who do not stutter?

• What types of anxiety and depressive symptoms are more likely to be associated with stuttering (if any)?

• Do symptoms of anxiety and depression increase with age in the stuttering group?

• Is there a relationship between stuttering severity and severity of anxiety/depression symptoms?

• Does this association vary depending on moderators such as sex, socio-economic status, family history, intervention receipt, or co-occurring language/cognitive deficits?

# Method

This systematic review follows the guidelines in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Statement (Liberati et al., 2009). The protocol was registered with PROSPERO (ID: CRD42019117327) in January 2019: https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=117327

## Measuring anxiety and depression in children

Rating scales and diagnostic interviews are commonly used to assess anxiety and depression symptoms and disorders in children and adolescents (see Thapar et al., 2015). Throughout this review, we refer to symptoms of anxiety rather than clinical diagnoses given the scales used in the included studies. Measuring symptoms of anxiety and depression in pre-schoolers is challenging and anxiety may be expressed differently by pre-school children relative to adults (Whalen et al., 2017). However, confirmatory factor analyses have demonstrated that anxiety symptoms in pre-school children align with anxiety disorder subtypes classified in the DSM-IV

(Spence et al. 2001), while the construct of preschool depression has been validated using developmentally appropriate assessment measures (Whalen et al., 2017).

## Conceptualising situation-specific and general anxiety

Anxiety self-report measures are heterogenous in so far as they likely tap different aspects of anxiety, therefore combining symptoms into a single construct of anxiety for the purposes of systematic review or meta-analysis can be problematic (Wall & Lee, 2021).

Therefore, we consider two separate anxiety domains based on the construct of anxiety that scales are considered to measure in the included studies:

- 'General anxiety' domain included scores obtained on both *trait anxiety* measures and *generalised anxiety* subscales. It has been suggested that generalised anxiety disorder is a manifestation of high trait anxiety (Rapee, 1991). The Revised Children's Manifest Anxiety Scales (RCMAS) and the Multidimensional Anxiety Scale for Children (MASC) are considered to measure chronic or trait anxiety, and total scores were therefore included in this domain (March et al., 1997; Reynolds, 1985).
- 'Situation-specific anxiety' domain comprised scores on both *state anxiety* measures and *social anxiety/phobia* subscales.

## Eligibility

*Inclusion criteria.* Studies were included if: (1) the primary focus was developmental stuttering; (2) participants were aged between two and 18 years[1] (3) study assessed symptoms associated

[1] Deviations from the protocol: the eligibility criteria originally specified a wider age range (2 – 25 years) for the purposes of consistency with the Special Educational Needs Disability (SEND) Code of Practice in England

with DSM-classified ‘anxiety disorders’ or ‘depressive disorders’; (4) anxiety and depression symptoms were measured using self- and parent-report symptom scales with acceptable reliability and validity; (5) study design included a non-stuttering comparison group; (6) report was published in English.

*Exclusion criteria.* Studies focusing on temperament were excluded. Although particular temperament traits may act as precursors to later onset of mental health conditions (Winter & Bienvenu, 2011), our research question focuses specifically on symptoms of anxiety and depression.

## Search Strategy

The first author conducted a literature search of databases, grey literature and a hand search of the Journal of Fluency Disorders and key reviews (Appendix A). Initially, all articles published prior to the end of January 2019 were included; a top-up search was conducted in January 2021. The first and second author screened study abstracts and titles using Distiller-SR software (Evidence Partners, Ottawa, Canada), and full texts were double screened for eligibility (inter-rater reliability, kappa = 0.89).

## Data Extraction

A coding scheme for extracting the relevant information about primary and additional outcomes was constructed and piloted (Higgins & Green, 2011) before two authors (RB and HH)

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(Children & Families Act, 2014) and internationally changing perceptions of adolescence. However, reviewers recommended a cut-off of 18 years. This resulted in the exclusion of three further studies.

independently extracted the data from included articles (inter-rater reliability kappa = 0.91). Three study authors were contacted for further information and two responded.

### *Primary outcome*

The primary outcome was the mean effect size difference (*Hedge's g*) in anxiety scores between the stuttering group and comparison group. The authors extracted the following information: (i) sample size (*n*); (ii) symptom measure and whether it purported to measure anxiety or depression; (iii) mean and standard deviation for anxiety and/or depression symptom scores for each group; (iv) between-group statistics comparing anxiety and depression symptoms.

### *Additional outcomes: moderators and study quality*

Additional information extracted for both groups for the purposes of moderator analyses and evaluation of study quality included: (i) age; (ii) sex; (iii) socio-economic background; (iv) family history of mental health concerns; (v) presence of co-occurring disorders; (vi) first language spoken; and (vii) respondent (child or parent).

Data extracted for the stuttering group only included: (i) method for confirming stuttering diagnosis; (ii) reported stuttering severity; (iii) family history of stuttering; (iv) receipt of speech or psychological intervention.

## Meta-Analysis Procedures

Effect sizes for each study were calculated based on the group mean, standard deviation and sample size using the *metafor* package (Viechtbauer, 2010) in *R* (R Core Team, 2020, version

4.0.4). The *escalc* function automatically corrects for positive bias when calculating the standardised mean difference, yielding a *Hedge's g* effect size (Viechtbauer, 2010). As the included studies varied in terms of sample size, the *Hedge's g* effect size was selected as it provides a more precise estimate when dealing with smaller samples (Hedges & Olkin, 1985). Similar to Cohen's d, *Hedge's g* effect sizes may be interpreted as small ($g < 0.30$), moderate ($g = 0.30 – g = 0.80$) or large ($g > 0.80$) (Cohen, 1988).

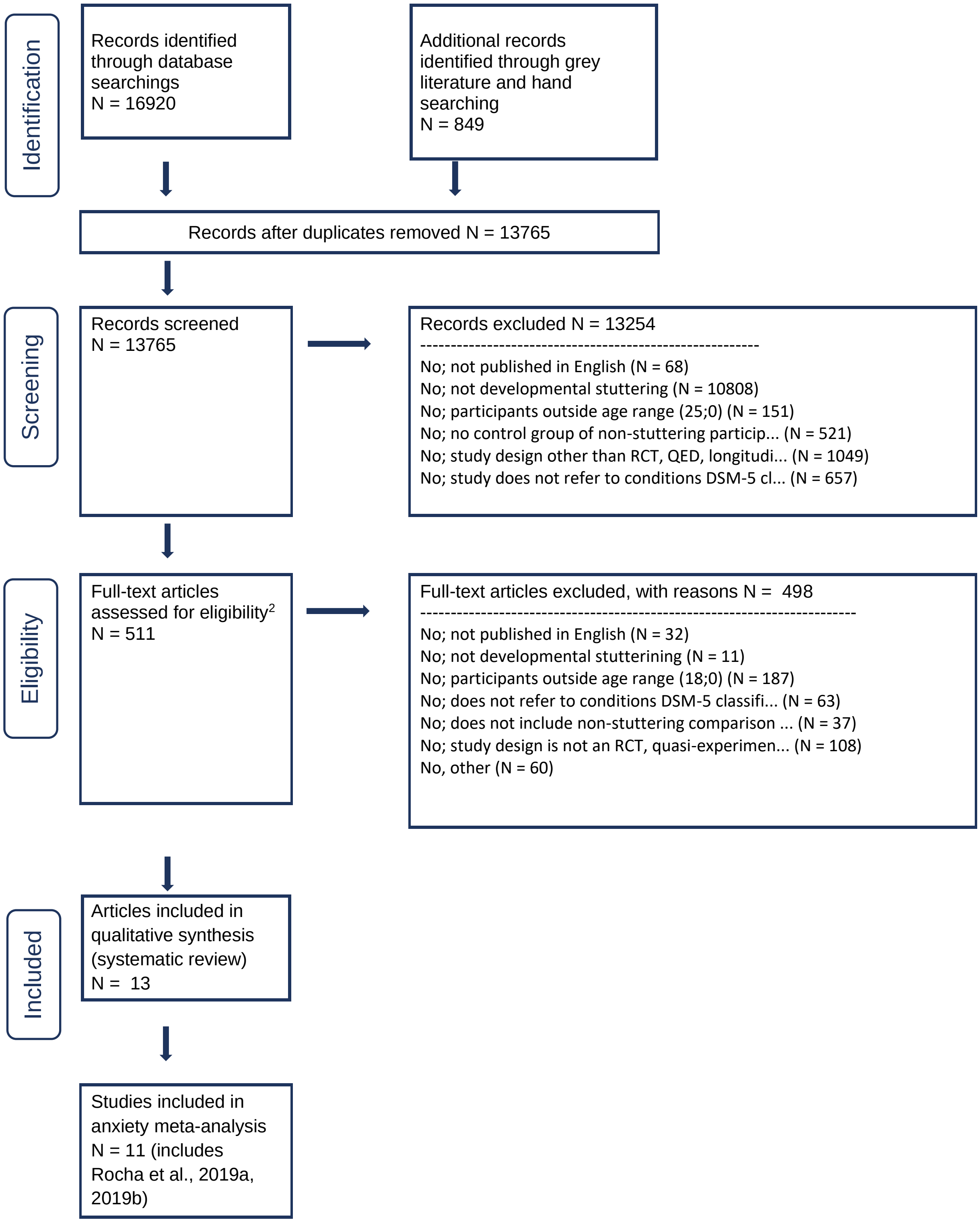
The systematic search identified 13 articles. One article (Rocha et al., 2019) reported data for two groups of participants and was treated as two studies in the analysis (Rocha et al., 2019a, Rocha et al., 2019b). Eleven studies were included in the quantitative analysis: eight reported anxiety symptoms and three reported anxiety and depression symptoms. One meta-analysis was performed using a random effects model as data came from different populations and there is variation across studies (Borenstein et al., 2009). There were too few studies to conduct a separate meta-analysis for depression symptoms (Borenstein et al., 2009). Several studies contributed more than one effect size as they reported multiple scores for the same sample, and consequently data were dependent. Robust Variance Estimation (RVE; Hedges, Tipton & Johnson, 2010) was used because the RVE method accounts for dependencies in the data (i.e. multiple scores from the same participants) when within-study covariance is unknown (Fisher & Tipton, 2015). This method for dealing with dependencies is increasingly being used in the fields of psychology, mental health and education, and was preferred over omitting datasets from the same study as it maximises use of available data and limits bias in decisions about which studies or measures to include (Tanner-Smith, Tipton & Polanin, 2016). The *robumeta* package (Fisher & Tipton, 2015) in R adjusts and weights studies appropriately by estimating correlations between measures from the same study sample. As the correlation coefficients were not known for those studies that provided multiple effect sizes, a conservative estimate was used (Rho = 0.8). A sensitivity analysis was conducted to determine whether the summary effect size was robust across different correlation levels. When conducting the analyses, the authors implemented the small-sample RVE estimators as described by Tipton (2015) to fit a correlated effects model with small sample corrections. If degrees of freedom (*df*) were less than four, the results were not considered reliable (Tipton, 2015; Fisher & Tipton, 2015). Meta-regression analyses were performed where possible to evaluate the effect of moderator variables on the summary effect size ($p < 0.05$). In cases where there were insufficient data in primary studies and/or too few studies ($k$<10) to perform the planned moderator analyses (Borenstein et al., 2009), qualitative report of extracted data is provided. Heterogeneity was quantified by calculating the Tau and $I^2$ statistics, neither of which are sensitive to the number of included studies (Borenstein et al., 2009). Tau describes the distribution of effect sizes around the mean effect, reflecting the amount of true heterogeneity (Borenstein et al., 2009). The $I^2$ statistic describes the proportion of the observed variability in effect estimates that is due to true heterogeneity rather than sampling error (Higgins et al., 2003; Borenstein et al., 2017). Low (25%), moderate (50%) and high (75%) values of $I^2$ have been tentatively suggested to aid interpretation (Higgins et al., 2003).

## Study Quality

Two authors independently assessed risk of bias in individual studies using a critical checklist (Appendix B; kappa = 0.85). Publication bias occurs when included studies are not representative, which leads to bias in the calculated effect size (Borenstein et al., 2009). To minimise publication bias and capture unpublished studies, grey literature was searched, such as reports and doctoral theses, and broad search terms used.

Evaluation of publication bias and small-study effects in the anxiety meta-analysis were conducted through observation of the funnel plot, Egger's Regression Test (Egger et al., 1997) and calculating the Failsafe N (Rosenthal, 1979). To account for dependencies, within-study effect sizes were aggregated using the *MAd* package (Del Re & Hoyt, 2018) in R, which implements Borenstein et al.'s (2009) procedure for aggregating dependent effect sizes (default $r$ = .50).

*[Figure 1: PRISMA flowchart outlining systematic review process (Distiller-SR)]*




[2] The 10 articles identified in top-up search (2021) added here

# Results

Following removal of duplicates, 13,765 references were identified, and 13,254 of these were excluded after title and abstract screening. After full text examination of the remaining 511 articles, a further 498 were excluded (Figure 1; see also *characteristics of excluded studies*, Supplementary Material). A total of 13 studies met our inclusion criteria, but three studies were excluded from the meta-analysis: two because they did not provide separate scores for anxiety and depression (Giorgetti et al., 2015; Tiğrak et al., 2020), and one reported incomparable group outcome data (proportion of children within each scoring band) (Andrews & Harris, 1964). Further information about these studies can be found in the Table 1.

*[Table 1: study characteristics (k = 13) for those that met inclusion criteria]*

## Study Characteristics

The 13 included studies comprised a total sample of 1, 165  participants, 541 of whom stuttered. Study sample sizes ranged from 14 to 225 participants, with ages ranging from three to 18 years (see Table 1).

*Table 1: Study characteristics (k=13) for those that met inclusion criteria*

| Study | Socio-economic Status (SES) | Co-occurring disorders | First language | Genetic factors (stuttering group) | Stuttering severity | Treatment status | Recruitment |
|---|---|---|---|---|---|---|---|
| ***Andrews & Harris (1964)** | No differences (Registrar General's classification) | Not reported | Not reported, UK study | Family history of stuttering (30/80)<br><br>Family history of mild (21/80) and severe (23/80) mental health problems | Graded on a 3-point scale: mild (56/80); moderate (18/80); severe (6/80) | 40% previously received treatment for stuttering | Community sample – recruited from schools in Newcastle-upon-Tyne, UK. |
| **Blood & Blood (2007)** | Middle to upper class (Hollingshead Four Factor Index, 1975) | Exclusion criteria references absence of physical or psychological disabilities | Not reported | Not reported | SSI-3: mild (11%); moderate (45%); severe (22%); very severe (22%) | Not stated. | SLT caseloads and advertisements in clinics. |
| **Blood et al. (2007)** | Middle to high class (Hollingshead Index of Social Position) | 44.4% had speech-language or non-speech-language disorder (stuttering group) | Not reported. US study | Not reported | SSI-3: mild (22.2%); moderate (27.7%); severe (22.2%); very severe (27.7%) | Only included participants who had received speech treatment | Contacted SLTs in public schools |
| **Craig & Hancock (1996)** | Not reported | Children assessed and those with language delays excluded (stuttering group) | Not reported Australian study | Not reported | Stuttering frequency (n): mild (44); moderate (29); severe (17) | All received treatment previously but no treatment three months | Recruited from those presenting for treatment |

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| | | | | | Reported correlation with state ($r$ = 0.115) & trait anxiety ($r$ = 0.045) | prior to the study | |
| **Davis, Shisca & Howell (2007)*** | Not reported | Children excluded based on school/parent report of language disorder (control group) | English as first language | Not reported | SSI-3: moderate (3); severe (11); very severe (4) | STAIC completed on average 29.44 months after 1-2 week intensive therapy course | Volunteer database |
| ***Giorgetti et al. (2015)** | Not reported | Exclusion criteria: psychiatric symptoms ro conditions, other relevant conditions | Brazilian Portuguese | Not reported | SSI-3: All classified at least mild | | Universidade Estadual Paulista |
| **Hollister (2015)** | American College Test (ACT) – mean parental education level 5 (bachelor degree). No significant difference between groups | Inclusion criteria: no neurological or intellectual impairment | English | Not reported | Iowa Scale: very mild (8); mild (8); mild-moderate (17); moderate (4); severe (5); very severe (4) | | Suburban & metropolitan areas in five geographical areas, USA. |

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| **Iverach et al. (2016)** | Middle income band: 34.7% (stuttering group); 35.3% (controls) | Stuttering group: current S&L difficulties (6.7%); Autism/ Asperger (2.7%); Autism + ADHD (1.3%).<br><br>Controls: S&L difficulties (0.7%); ADHD (3.3%); Autism/ Asperger (2.7%) | Main language English: 96% (stuttering group) and 95.3% controls | Positive family history of stuttering: 56% | Parent/child report: mild (31%); moderate (52%); severe (17%) | 80% currently enrolled in therapy.<br><br>78.7% previously accessed therapy for stuttering; 28% sought mental health assessment / treatment | Via speech pathology clinics and advertisements in general community. |
| **Mulcahy et al. (2008)** | Not reported | Not reported<br><br>No history of pharmacological intervention for anxiety-related disorders | Not reported Australian study | Not reported | %SS: mild (12%); moderate (47%); severe (41%)<br><br>Reported correlation with state ($r$ = 0.04) & trait anxiety ($r$ = 0.09) | 68% currently enrolled in speech therapy. All had previously accessed therapy for stuttering. | Via clinic waiting lists and clinicians in Western Australia. |
| **Natarelli (2018)** | Hollingshead Four Factor Index of Social Status (1975).<br><br>Significant between group differences. Stuttering group classified as medium-high SES | Exclusion criteria: no current or past psychiatric disorders; no use of psychotropic medications. | Functional written and spoken Italian | Not reported | SSI-4: very mild (10), mild (6), moderate (3).<br><br>No significant correlation between RCMAS Total Score and SSI-4 ($r$ = -.09) | Majority enrolled in speech therapy (none within last 6-months).<br><br>Three never received treatment | SLTs & Psychologists identified potential families |

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| **Rocha et al. (2019)** | Parent education (most reported min. graduate level of education) | Inclusion criteria: absence of neurological, psychiatric and learning disorder | Monolingual Portuguese speakers | 60% had positive family history | SSI-4 used to confirm & diagnose stuttering | Speech therapy during (22%) or prior to (22%) study. 28% never received treatment. | Via SLTs and school teachers who referred eligible families. |
| ***Tiğrak et al. (2020)** | Maternal education | Inclusion criteria: absence of neurological, psychological or developmental problems.<br><br>Assessed receptive & expressive language skills. | Native Turkish speakers | Inclusion criteria: no parent history of speech & language difficulties | 3+ stuttering incidents per 100 words. | Inclusion criteria: no history of speech therapy | Selected from applications to clinic. |
| **van der Merwe et al. (2011)** | Not reported | Age appropriate speech & language skills in both groups following assessment | Not reported New Zealand study | Not reported | Stutter-like disfluencies (SLDs): ranged from 3% - 24%<br><br>Reported correlation with total anxiety ($r$ = 0.13); social phobia ($r$ = -0.02); generalised anxiety ($r$ = 0.48) | 4/7 children currently enrolled, two awaiting therapy. | Speech & Hearing Clinic – identified due to involvement in therapy or parental concern. |

## Anxiety symptoms meta-analysis

Eleven studies (*n* = 851 participants, comprising 384 participants who stutter) contributing 26 effect sizes were analysed. Rocha et al. (2019) divided their sample into ‘younger’ and ‘older’ age groups, which were entered as separate studies in the model as they were different groups of participants. A significant, moderate summary effect size was obtained for anxiety symptoms ($g$ = 0.42, $p$ = 0.02, 95% CI [0.1, 0.743], $df$ = 9.45), indicating higher mean anxiety symptom scores were detected in children and adolescents who stutter relative to fluent peers. Results indicated considerable heterogeneity ($I^2$ = 72.8% ; Tau =0.40 ) across included studies.

The individual effect sizes obtained for between-group comparisons are provided in Table 2. Negative effect sizes were obtained in three studies, suggesting the comparison group scored higher or equal to the stuttering group on measures of anxiety. However, small to moderate effect size differences were obtained in most studies.

Sensitivity analyses were conducted by removing one study (all effect sizes) at a time and re-running the random effects model. The summary effect size ranged from $g$ = 0.33 to $g$ = 0.50, and all remained statistically significant at $p < 0.05$. The authors also performed a random effects meta-analysis using aggregated effect sizes for each study (11 effect sizes). The *MAd* package in R was used to aggregate effect sizes while accounting for dependencies. The summary effect remained reasonably consistent with the summary effect calculated using the RVE method ($g$ = 0.42, p = 0.004, 95% CI [0.14, 0.71], $I^2$ = 78.04%, Tau = 0.41).

*Table 2: Studies included in the anxiety meta-analysis, organised in approximate order of effect size*

| **Author** | **Stutter group (n, sex, age)** | **Non-stutter group (n, sex, age)** | **Anxiety measure** | **Respondent** | **Country** | **Anxiety domain** | **Effect Size *(g)* [95% CI]** |
|---|---|---|---|---|---|---|---|
| Craig & Hancock (1996)* | 96<br>78M : 18F<br>9–14yrs | 104<br>59M : 45F<br>9–14yrs | State-Trait Anxiety Inventory for Children (STAIC) | Self-report | Australia | GENERAL<br>*Trait anxiety measure* | -0.15<br>[-0.43 – 0.13] |
| Van der Merwe et al. (2011) | 7<br>5M : 2F<br>3;3 – 4;11yrs | 7<br>5M : 2F<br>3;2 – 4;10yrs | Pre-school Anxiety Scale (PAS) | Parent-report | New Zealand | *PAS total score* | -0.06<br>[-1.11 – 0.98] |
| | | | | | | GENERAL<br>*Generalised anxiety disorder subscale* | -0.27<br>[-1.32 – 0.79] |
| | | | | | | SITUATION-SPECIFIC<br>*Social phobia subscale* | 0.00<br>[-1.05 – 1.05] |
| Rocha, Yaruss & Rato (2019a) | 31<br>25M : 6F<br>7–9yrs | 31<br>15M : 16F<br>7–9yrs | Multidimensional Anxiety Scale for Children (MASC)[1]<br>*Portuguese version* | Self-report | Portugal | GENERAL<br>*MASC total score* | -0.06<br>[-0.55 – 0.44] |
| | | | | | | SITUATION-SPECIFIC<br>*Social anxiety subscale* | 0.13<br>[-0.37 – 0.62] |
| Rocha, Yaruss & Rato (2019b) | 19<br>11M : 8F<br>10–12yrs | 19<br>7M : 12F<br>10–12yrs | Multidimensional Anxiety Scale for Children (MASC)[1]<br>*Portuguese version* | Self-report | Portugal | GENERAL<br>*MASC total score* | 0.15<br>[-0.49 – 0.79] |
| | | | | | | SITUATION-SPECIFIC | -0.20 |

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|---|---|---|---|---|---|---|---|
| | | | | | | *Social anxiety subscale* | [-0.84 – 0.43] |
| Hollister (2015) | 46<br>36M : 10F<br>8–15yrs | 46<br>36M : 10F<br>8–15yrs | The MacArthur Health & Behavioural Questionnaire 2.1 (HBQ 2.1) | Parent-report | USA | SITUATION-SPECIFIC<br>*Social anxiety subscale* | 0.26<br>[-0.15 – 0.68] |
| | | | | | | GENERAL<br>*General anxiety subscale* | 0.54<br>[0.12 – 0.95] |
| Natarelli (2018) | 19<br>14M : 5F<br>11–14yrs | 19<br>Matched by sex, grade, ethnicity<br>11–14yrs | Revised Children's Manifest Anxiety Scale - Second Edition (RCMAS-2)<br>*Italian version* | Self-report | Italy | GENERAL<br>*RCMAS Total score* | 0.38<br>[-0.27 – 1.02] |
| Iverach et al. (2016) | 75<br>63M : 12F<br>7–12yrs | 150<br>126M : 24F<br>7–12yrs | Spence Children's Anxiety Scale (SCAS) | Child-report (SCAS-C)<br><br>Parent-report (SCAS-P) | Australia | *SCAS total score* | SCAS-P: 0.51<br>[0.22 – 0.79]<br><br>SCAS-C: 0.46<br>[0.18 – 0.74] |
| | | | | | | GENERAL<br>*Generalised anxiety subscale* | SCAS-P: 0.48<br>[0.20 – 0.76]<br><br>SCAS-C: 0.32<br>[0.04 – 0.60] |
| | | | | | | SITUATION-SPECIFIC<br>*Social anxiety subscale* | SCAS-P: 0.57<br>[0.29 – 0.85]<br><br>SCAS-C: 0.56<br>[0.28 – 0.84] |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| Davis et al. (2007) | 18<br>16 M : 2F<br>10–16yrs | 19<br>14M : 5F<br>10–15yrs | State-Trait Anxiety Inventory for Children (STAIC) | Self-report | England | GENERAL<br>*Trait anxiety measure* | 0.25<br>[-0.40 – 0.90] |
| | | | | | | SITUATION-SPECIFIC<br>*State anxiety measures* | Friends: 0.43<br>[-0.22 – 1.08]<br><br>Phone: 0.69<br>[0.03 – 1.36]<br><br>Class: 0.86<br>[0.19 – 1.53]<br><br>Shop: 1.03<br>[0.35 – 1.72] |
| Blood & Blood (2007) | 18<br>Male<br>11–12yrs | 18<br>Male<br>Age matched | Revised Children's Manifest Anxiety Scale (RCMAS)[1] | Self-report | USA | GENERAL<br>*Total score* | 0.71<br>[0.04 – 1.38] |
| Blood et al. (2007) | 36<br>30M : 6F<br>12;8 – 18;7yrs | 36<br>Age & gender matched | Revised Children's Manifest Anxiety Scale (RCMAS)[1] | Self-report | USA | GENERAL<br>*Total score* | 0.86<br>[0.38 – 1.35] |
| | | | | | | | |
| Mulcahy et al. (2008) | 19<br>18M : 1F<br>11–18yrs | 18<br>16M : 2F<br>12–17yrs | State and Trait Anxiety Inventory (STAI) | Self-report | Australia | GENERAL<br>*Trait anxiety measure* | 2.00<br>[1.21 – 2.79] |
| | | | | | | SITUATION-SPECIFIC<br>*State anxiety measure* | 1.18<br>[0.48 –1.88] |

### *Meta-regression: situation-specific and general anxiety domains*

This review also considered the extent to which higher anxiety is more likely to be related to social and communicative situations or general anxiety in children and adolescents who stutter. Data were classified into two anxiety domains: 11 effect sizes were included in the situation-specific domain and the general anxiety domain comprised 12 effect sizes (see Table 2).

Meta-regression analyses indicated a moderate effect size difference for both the situation-specific ($g$ = 0.42, 95% CI [-0.01, 0.85], $df$ = 5.36, $p$ = 0.06) and general anxiety ($g$ = 0.44, 95% CI [0.01, 0.86], $df$ = 7.46, $p$ = 0.05) domains, although only the general anxiety domain was significant, with wide confidence intervals for both situation-specific and general anxiety. The results of the meta-regression analysis would tentatively suggest that elevated anxiety is observed in measures of both social and general anxiety, however it may be that the measures used were not sufficiently sensitive to distinguish between the two.

**Situation-specific Anxiety.** Estimates of situation-specific anxiety were based on five state anxiety[3] and six social anxiety[4] subscale scores. Small to moderate effect size differences were obtained for most studies; the largest effect sizes were observed in measures of state anxiety (Davis et al., 2007; Mulcahy et al., 2008). When considering the mean scores in individual studies, Davis et al. (2007) reported significantly higher mean scores for three of four states in the persistent stuttering group compared to controls. Craig and Hancock (1996) found the mean state anxiety score for the stuttering group was lower than the normative sample. Social anxiety/phobia subscale group scores, both child- and parent-reported, differed significantly in

[3] Four of these state anxiety scores were from the same sample (Davis et al., 2007)
[4] Includes both child- and parent-scores on the social anxiety subscale (Iverach et al., 2016)

only one study (Iverach et al., 2016) of those measuring social anxiety. Although group differences did not reach statistical significance, higher mean scores were reported for the stuttering group in two other studies (Hollister, 2015; Rocha et al., 2019a). The Iverach et al. (2016) study included the largest sample (*n* = 225) and it may be that the smaller studies did not have sufficient power to detect differences.

**General Anxiety.** Five studies reported significant differences in mean group scores (Blood & Blood, 2007; Blood et al., 2007; Hollister, 2015; Mulcahy et al., 2008; Iverach et al. 2016). Three further studies (Iverach et al., 2016 – child report; Natarelli, 2019, Rocha et al., 2019b) reported higher mean scores for the stuttering group, although differences did not reach significance. The summary effect size for general anxiety was based on total scores obtained from the Revised Children's Manifest Anxiety Scale (RCMAS; Blood & Blood, 2007; Blood et al., 2007; Natarelli, 2018) and the Manifest Anxiety Scale for Children (MASC; Rocha et al, 2019a; Rocha et al, 2019b); generalised anxiety subscale scores; and trait anxiety scores (State-Trait Anxiety Inventory). Small effect sizes were obtained for most studies, with three studies characterised by large effect sizes (Blood & Blood, 2007; Blood et al., 2007; Mulcahy et al., 2008).

### *Clinical and subclinical anxiety*

For the most part, studies reported mean differences in symptom summary scores, rather than the extent to which participants met clinical thresholds for anxiety disorder. Elevated anxiety scores do not necessarily mean that an individual has a clinical anxiety disorder. Two studies reported that mean scores fell within the normative range (Blood et al., 2007; Iverach et al., 2016), though

Blood and Blood (2007) reported 39% of the stuttering group scored at least one standard deviation above the normative mean.

One study reported rates of clinical and subclinical anxiety disorder based on a structured diagnostic interview (Youth Online Diagnostic Assessment; YODA). Iverach et al. (2016) found 24% of participants who stutter met criteria for social anxiety disorder compared to 5% of the non-stuttering group. While there were no significant differences in rates of clinical generalised anxiety disorder, rates of subclinical generalised anxiety disorder were significantly higher in the stuttering group.

## *Moderator Analyses*

Data about potential moderators were extracted from all anxiety studies included in the meta-analysis to examine the relationship with the primary outcome.

**Age**. Only one preschool study was included in the meta-analysis and no significant group differences were reported for any of the anxiety subscales (van der Merwe et al., 2008). Removal of this sole pre-school study did not meaningfully change the summary effect ($g$ = 0.45, 95%CI [0.11, 0.79], $df$ = 8.7, $p$ = 0.02). Rocha et al. (2019) reported no significant group differences in mean anxiety score in the younger (age 7–9 years) or older (10–12 year) age groups.

**Sex**. It was not possible to examine the effects of sex as the majority of studies did not report mean scores separately by sex. However, Craig and Hancock (1996) reported no significant association between sex and state or trait anxiety scores.

**Socio-economic group (SES).** None of the studies analysed scores by SES. Seven of the included studies reported SES, measured as parental education level, occupation and/or income. Two studies explicitly state the sample to be middle class and above (Blood & Blood, 2007; Blood et al., 2007). Most parents in the Rocha et al. (2020) study were graduates, while the mean education level in Hollister (2015) was bachelor's degree, both suggesting a middle to high SES. Only two studies (Iverach et al., 2016; Natarelli, 2018) reported significant differences in measures of SES between stuttering and non-stuttering groups. Iverach et al. (2016) found significantly higher parental education level for the non-stuttering group, although groups did not differ on reported occupation or income. Natarelli (2018) found the stuttering group were more likely to be categorized as medium-high SES than the non-stuttering group (low SES).

**Co-occurring disorders.** Blood et al. (2007) reported adolescents who stutter with co-occurring disorders (n = 16) had higher levels of anxiety (Cohen's $d = 1.4$, $p < 0.001$) than those without co-occurring difficulties (n = 20). Two studies confirmed presence or absence of co-occurring speech and/or language disorders with standardised assessments and subsequently included only those children with age-appropriate language skills (Craig & Hancock, 1996; van der Merwe et al., 2008). Neither of these studies found significant differences in anxiety scores between stuttering and non-stuttering groups.

**Respondent**. Symptom severity on mental health measures can vary considerably depending on respondent (De Los Reyes & Kazdin, 2005). In the present review, most studies ($n = 8$) utilised self-report measures; only two involved parent-reported symptoms. One study compared parent-reported and child-reported scores on the same scale, and found that parent mean scores were significantly lower than child mean scores on SCAS Total and subscale scores (Iverach et al., 2016).

**First language.** Anxiety symptoms not reported in relation to first language.

**Family history of stuttering and/or mental health**. Anxiety symptoms were not reported in relation to family history of stuttering or mental health.

**Receipt of intervention.** Included studies did not report anxiety scores separately for participants who had or had not received speech or psychological intervention.

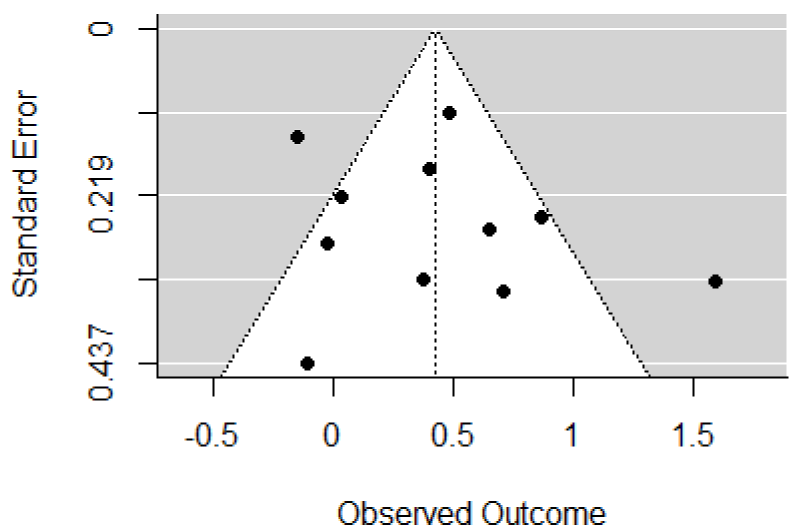
**Stuttering severity.** Based on the information available in included studies, it was not possible to analyse the effect of stuttering severity on primary outcomes. However, five studies reported that severity of stuttering was not associated with anxiety scores (Blood et al., 2007; Craig & Hancock, 1996; Mulcahy et al., 2008; Natarelli, 2018; van der Merwe et al., 2011). This suggests that stuttering severity is not necessarily associated with anxiety in childhood, which contrasts with some of the adult literature (Ezrati-Vinacour & Levin, 2004).

Limited data on demographic variables restricted the extent to which conclusions could be drawn regarding the effect of moderator variables on anxiety levels, although stuttering severity was found not to be associated with anxiety score in those studies that reported it. Future research should consider multiple factors in assessment of anxiety in stuttering to elucidate variable outcomes.

## *Small study and publication bias*

We investigated potential bias using funnel plots, Egger's Regression Test and the Failsafe N. Observation of the funnel plot (Figure 2) suggests some asymmetry given the absence of studies in the lower right-hand corner of the plot and the fact that several observations fall outside the 95% confidence interval, which is indicative of between-study heterogeneity (Sterne & Harbord, 2004). However, asymmetry in funnel plots may not relate to publication bias but other study factors (Sutton, 2009). Egger's Regression Test was not significant, which is consistent with funnel plot symmetry ($z = 0.69$, $p = 0.49$). The Failsafe N indicated that 129 additional studies would be necessary to yield a non-significant summary effect, which indicates relative robustness to publication bias (Becker, 2005).

*Figure 2: funnel plot of included studies*



## Depression symptoms qualitative analysis

Three studies (five effect sizes) contributed depression symptom scores for 355 participants, of whom 140 stutter. Sample sizes ranged from 38 to 225 participants (Table 3).

Higher mean depression scores were observed for the stuttering group across all symptom measures in included studies, except for the mother-reported symptoms on the Depression Anxiety in Youth Scale (DAYS; Natarelli, 2018). However, none reached conventional levels of statistical significance. The age of onset for depression is typically later than for anxiety and risk increases substantially during mid to late adolescence, with some arguing particular vulnerability from age 15 years (Hankin et al., 1998; Kessler et al., 2005; Lewinsohn et al., 1994). These three studies reported both depression and anxiety scores for the sample (Hollister, 2015; Iverach et al., 2016; Natarelli, 2018). Despite the comorbidity between anxiety and depression, present findings indicate anxiety to be of greater concern than depression for children who stutter, which may be partly attributable to the upper age limit in these three studies relative to the average age of onset of depression reported in the literature. Due to the small number of datasets and the absence of information for moderator variables defined in the protocol, no further analyses were conducted. These findings are consistent with the previous qualitative review on this subject, and contribute evidence from a further three studies that include a non-stuttering comparison group.

*Table 3: Effect sizes calculated for depression studies included in the review, arranged in order of effect size magnitude.*

| **Author** | **Stutter group (n)** | **Non-stutter group (n)** | **Depression measure** | **Respondent** | **Country** | **Effect size (*g*) [95% CI]** |
|---|---|---|---|---|---|---|
| Natarelli (2018) | 19<br>14M : 5F<br>11 – 14yrs | 19<br>Matched for sex, ethnicity & grade<br>11 – 14yrs | Depression Anxiety in Youth Scale (DAYS)* | Parent-report | Italy | Mother-reported:<br>0.0<br>[-0.64 – 0.64]<br><br>Father-reported:<br>0.18<br>[-0.46 – 0.82] |
| Hollister (2015) | 46<br>36M : 10F<br>8 – 15yrs | 46<br>36M : 10F<br>8 – 15yrs | The Early Adolescence Temperament Questionnaire-Revised (EATQ-R) | Parent-report | USA | 0.28<br>[-0.13 – 0.69] |
| | | | The MacArthur Health and Behavioural Questionnaire 2.1 (HBQ-2.1) | Parent-report | USA | 0.31<br>[-0.11 – 0.72] |
| Iverach et al. (2016) | 75<br>63M : 12F<br>7 – 12yrs | 150<br>126M : 24F<br>7 – 12yrs | Short Moods & Feelings Questionnaire (SMFQ) | Self-report | Australia | 0.34<br>[0.06 – 0.61] |

*N = sample size, sex ratio and age range (years), male : female ratio; depression measure used in study; self- or parent-report measure; country in which study was undertaken; effect size (Hedge's g) and confidence intervals .*
*Authors extracted depression scores only as poor internal consistency was reported for anxiety subscale (Natarelli, 2018, p91). Separate mother- and father-reported scores provided.

## Study Quality

Risk of bias was assessed in the 13 studies using an adapted version of the Critical Checklist for cohort studies (Appendix B), and was judged unclear for all studies.

### *Selection bias*

Ten studies recruited from clinic waiting and caseload lists, or via health and education professionals. One study recruited from a volunteer database, which may have resulted in self-selection bias. Only two studies comprised a representative community sample (Andrews &

Harris, 1964; Hollister, 2015). Neither study reported significant differences between groups on most anxiety and depression measures, although Hollister (2015) reported higher scores on the generalised anxiety subscale in the stuttering group relative to peers.

### *Power analysis*

It has been suggested that research into the relationship between stuttering and anxiety has been limited by small sample sizes and insufficient power, which may contribute to the variable findings in the literature (Craig, 1990; Menzies et al., 1999). One included study (Iverach et al., 2016) conducted statistical power analysis to determine the appropriate sample size to achieve adequate power for detecting the effect of interest. The other included studies did not report whether they had conducted power analyses.

### *Mental health status of participants*

Seven studies controlled for current mental health problems in their study samples by stipulating absence of psychiatric conditions or medications in the eligibility criteria. One study (Iverach et al., 2016) reported the proportion of the sample with a current psychiatric diagnosis and/or medication, while Andrews and Harris (1964) stated the proportion with a family history of mental health problems. Neither study reported the group scores separately for affected versus non-affected participants.

### *Access to speech or psychological intervention*

Nine studies reported participants had received previous or current speech and language intervention but most did not describe the intervention approach. Only one study reported prior access to mental health services (Iverach et al., 2016). Most study samples combined participants who had and had not received speech and language therapy, though Tiğrak et al. (2020) excluded participants with a history of speech therapy. Two studies reported that the stuttering group were not receiving speech and/or psychological therapy for stuttering at the time of the study nor in the months leading up to it (Craig & Hancock, 1996; Natarelli, 2018). None of the studies reported anxiety/depression symptoms separately according to intervention status.

## Discussion

The aim of the present review was to investigate whether children and adolescents who stutter present with elevated symptoms of anxiety or depression relative to non-stuttering peers. The major finding from the meta-analysis is that there is a moderate summary effect size difference indicating increased anxiety symptoms in some children and adolescents (aged 3 to 18 years) who stutter relative to fluent peers. In the one study that employed diagnostic interviews, approximately 1/3 of participants who stutter met criteria for anxiety disorder (Iverach et al., 2016).

Only three studies reported symptoms of depression. The small number of studies means that we could not reliably estimate an overall effect size. While mean depression scores for the stuttering group were higher than the comparison group for four of the five measures reported across studies, this difference did not reach the threshold for statistical significance in any study. This may relate to the age range of the sample (<15 years) given later onset of depression in the general population.

On balance, this evidence suggests a need to carefully monitor anxiety symptoms in young people who stutter and highlights a need for further research into depression. Enhanced reporting of broader participant characteristics in empirical studies of stuttering could elucidate risk and protective factors for anxiety and depression in this population.

## Anxiety symptom profiles associated with stuttering

This review also aimed to investigate whether elevated anxiety symptoms are associated to a greater extent with situations that place emphasis on social interaction and communication. Previous research suggested that elevated anxiety in adults who stutter occurs in specific social situations, and thus has been considered an expected or rational response to the experience of stuttering (Diehl et al., 2019; Ezrati-Vinacour & Levin, 2004; Messenger et al., 2004; Miller & Watson, 1992; Vanryckeghem et al., 2017). In the present review, moderate effect sizes were evident for both situation-specific and general anxiety domains, although the wide confidence intervals suggest considerable variation in anxiety. Interestingly, Davis et al. (2007) reported significant differences between the persistent stuttering and control groups in all state anxiety measures except for 'talking with friends', which may suggest that individuals felt less anxious with a familiar listener. The present analyses cannot determine whether elevated anxiety symptoms are exclusively associated with social situations, and instead longitudinal studies are necessary to ascertain how anxiety symptoms may develop over time.

### *Assessment of anxiety in the literature*

All included studies utilised scales that are widely-used for clinical and research purposes, and had acceptable psychometric properties. However, anxiety symptom scores combined in the meta-analysis were obtained from seven different symptom scales; the State-Trait Anxiety Inventory (STAI) and the Revised Children's Manifest Anxiety Scale (RCMAS) were the only measures employed in more than one study. Scales tap different dimensions of anxiety and therefore cannot necessarily be considered interchangeable with one another (Wall & Lee, 2021; Keedwell & Snaith, 1996).  For example, the State-Trait Anxiety Inventory (STAI; Spielberger, 1983), RCMAS (Reynolds & Richmond, 2002) and Multidimensional Anxiety Scale for

Children (MASC; March et al., 1997) primarily assess the constructs of state and/or trait anxiety. Equally, some rating scales assess symptoms broadly in line with specific anxiety disorders and map onto DSM-criteria: Spence Children's Anxiety Scale (SCAS; Spence, 1998); Pre-school Anxiety Scale (PAS; Spence & Rapee, 1999); and McArthur Health Behaviour Questionnaire 2.1 (HBQ-2.1; Armstrong et al., 2003). The extent to which mean scores differed between stuttering and non-stuttering groups may in part reflect the multitude of symptoms and aspects of anxiety that scales assess. Furthermore, existing symptom-report scales may have poor sensitivity for detecting anxiety associated with the specific experiences of stuttering (Veerabhadrappa et al., 2021). A recent systematic review found insufficient availability of measures for robustly assessing speech-related anxiety in children who stutter (Jones et al., 2021). Future research could consider using tools that are sensitive to the experiences of this clinical population to improve understanding of risk and resilience in anxiety.

## Changing symptom profiles with age

Higher rates of anxiety, and to a lesser extent depression symptoms, have been observed in adults who stutter (Craig & Tran, 2014; Livingtsone-Pountney & Mitrevski, 2019). Previous review of the literature could not determine the age of anxiety onset in children who stutter, concluding that anxiety may increase over time (Smith et al., 2014). Determining the approximate onset of symptoms would be beneficial to the management and possible prevention in this clinical population.

In the present meta-analysis, most anxiety studies involved school-aged children and adolescents (7-18 years), while removal of the pre-school study made little difference to the summary effect size. This indicates an association between stuttering and elevated anxiety symptoms may be apparent in children of primary school-age. Recently, Veerabhadrappa et al. (2021) concluded that speech-related anxiety could be present in children who stutter from seven years of age.

Rocha et al. (2019) reported no significant age effects in mean anxiety scores when comparing younger or older samples, whereas Tiğrak et al. (2020) found the stuttering group had significantly higher anxiety/depression scores compared to controls in each age group assessed (early childhood, middle childhood, adolescence). This disparity may be partly explained by the scales used as Tiğrak et al. (2020) compared groups on the 'anxious/depressed' subscale of the Child Behaviour Checklist (CBL), whereas groups were compared on the MASC in the Rocha et al. (2019) study. The two groups were also much closer in age in the Rocha et al. (2019) study. As these were cross-sectional designs, it is difficult to infer the extent to which age moderates risk of anxiety within an individual. Instead longitudinal designs could inform the psychosocial development of children over time, informing our understanding of the age at which children may be more vulnerable to anxiety and the potential risk and protective factors involved.

## Factors moderating the association between stuttering and anxiety and depression

As can be seen in the present review, and the adult literature, not all individuals who stutter present with heightened anxiety. One explanation for such variability between studies could be that other factors moderate the association between stuttering and elevated anxiety and possible depression, which increases risk for some and serves to play a protective role for others.
Many of the studies in the current review were characterised by relatively small samples and few reported sufficient clinical or demographic information for in-depth analyses of potential moderating factors. While higher anxiety scores have been observed in children who stutter with

additional communication disorders (Alm, 2014; Blood et al., 2007; Smith et al., 2017), there was insufficient data to analyse the extent to which co-occurring disorders may be involved in any association between anxiety and stuttering in children in this review. In future, studies investigating mental health with this population could report child, genetic, broader family or parent characteristics (Park et al., 2021), and environmental/social variables that may act as additional risk or protective factors for anxiety and depression.

Another contributing factor to variable research findings may relate to ascertainment, which has been cited in published reviews (Iverach et al., 2011; Menzies et al., 1999; Smith et al., 2014). Children and adolescents who stutter are likely to have accessed speech-language pathology services, where treatment options may focus on developing fluency, or on psychological approaches to managing dysfluency (see Baxter et al., 2016). Over half of the studies included in this meta-analysis were at risk of recruitment bias and reliance on clinically ascertained cohorts. This is methodologically problematic because young people may access services if they are anxious about communication or distressed by their fluency, thus elevating anxiety symptoms (Craig et al., 2003). On the other hand, clinically referred cohorts are likely to be receiving treatment for stuttering, which may itself influence anxiety levels (Craig, 1994). Population cohorts are therefore needed to generate unbiased estimates of anxiety and depression, and to potentially elucidate factors associated with resilience in this population.

## Limitations

Our conclusions are limited by the small number of studies, small sample sizes within some studies, and between-study variation. Exclusion of studies that did not include a non-stuttering comparison group (e.g. Gunn et al., 2014; Smith et al., 2017) limited the number of studies that contributed to the overall effect size but a comparison group was necessary to estimate the relevant effect size. Although we aimed to assess anxiety and depression across a broad developmental period, we only included one pre-school study, as most studies at this age measured temperament as a precursor to anxiety or depression. Future longitudinal studies are essential to inform the onset and trajectory of mental health outcomes in the stuttering population.

Anxiety is a complex construct that is measured in a myriad of ways. Given the limited number of studies available and the variety of symptom measures relied upon, our meta-analysis collapsed data from scales that purported to measure different anxiety constructs and symptoms, and therefore were not necessarily 'capturing the same "anxiety"'(Wall & Lee, 2021, p. 16). We also grouped measures of social and state anxiety, as potentially different to measures of general and trait (situation invariant) anxiety; however, some readers may not consider social anxiety to be a transitory state. State and trait anxiety can be seen as intertwined, for instance levels of state anxiety are the result of both the person (trait) and the situation (Endler & Kocovski, 2001), yet we chose to group them separately. Consequently, we acknowledge that other researchers may choose to group these scales differently.

## Clinical implications

Our findings suggest that some children and adolescents who stutter experience greater symptoms of anxiety than peers, and may have a tendency towards increased depressive symptoms, though this does not necessarily mean that individuals meet clinical thresholds for anxiety disorder or clinical depression. It must be recognised that children who stutter are not a homogenous group and consequently not all children and adolescents who stutter present with anxiety symptoms. Furthermore, the present analysis cannot determine any causal relationship between anxiety and stuttering. Nevertheless, these findings are important for alerting professionals and parents of the need to support the well-being of children who stutter.

The present review also illustrates the need to attenuate the risk of developing anxiety and depression in children and adolescents who stutter, especially given evidence that poor mental health may be associated with poorer treatment outcomes in adults who stutter (Iverach et al., 2009b). Interventions may seek to reduce anxiety and foster resilience in children receiving speech and language therapy for stuttering. For instance, introducing a resilience component to stuttering therapy improved fluency, emotional, behavioural and resilience outcomes in pre-school children who stuttered (Druker, Mazzucchelli & Beilby, 2019). Consequently, the present review highlights the importance of early identification, on-going monitoring of psychosocial development, and consideration of onward multi-disciplinary referral in the management of children who stutter.

## Conclusion

Meta-analysis conducted with 11 studies indicates that children and adolescents who stutter have, on average, greater symptoms of anxiety relative to peers who do not stutter. Variability across studies likely reflects differences in choice of anxiety scales, participant treatment status and moderating factors, such as participant age and presence of co-occurring disorders. There were too few studies to draw robust conclusions about risk of depression in this population. However, these preliminary findings, coupled with recognition of the comorbidity between anxiety and depression in the general population, warrant further research in this area.

Longitudinal studies that assess anxiety and depression symptomology throughout childhood and adolescence will be critical. Future studies should also consider the factors that may moderate the development of anxiety and depression in order to identify additional malleable targets for improving the mental well-being of young people who stutter.

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## Appendix A: search strategy

*Screening process and data extraction was assisted through the use of forms created by the authors in Distiller-SR, based on templates provided.*

**Database Search Strategy**

The following databases were included in the literature search: Health & Psychosocial Instruments (HAPI); MEDLINE; PsycINFO; PsychTESTS; PubMed; ERIC; CINAHL; Web of Science core collection; ASSIA: Applied Social Science Index and Abstracts; AMED (Allied & Complimentary Medicine); IBSS: International Bibliography of Social Sciences; Cochrane Central Register of Controlled Trials (CENTRAL); Scopus; LLBA.

All database and grey literature searches used the following search term formula:

(1) 'disorder' AND (2) 'mental health scope': *stutter* OR stutter* OR 'fluency disorder*' OR dysfluen* AND 'mental health' OR 'mental health difficult*' OR 'mental health disorder*' OR anxiety OR 'anxiety disorder*' OR depression OR 'clinical depression' OR 'social phobia*.*

The authors also completed a hand search of the Journal of Fluency Disorders search terms: '*Stutter*' AND 'anxiety' OR 'depression'.*

**Grey literature search strategy**

Grey literature coding system: '*searched; nothing found*', '*not searched; not relevant', 'searched; results found*', '*results may be of peripheral interest*'. The references that were coded '*results may be of peripheral interest'* were uploaded into a separate EndNote file.

**Top-up search (February 2021)**

Authors re-ran the database search to include any studies published between January 2019 and January 2021. Searched: Journal of Fluency Disorders, Google Scholar, UCL library explore function and PubMed.

Simplified search terms: stutter or stammer AND mental health OR anxiety OR depression

## Appendix B: Risk of Bias

*Table below includes the number of studies considered to be of low/unclear/high risk of bias for the stated parameters (k=13).*

| Question | Low risk % (no. of studies) | Unclear risk (probably yes) | Unclear risk (probably no) | High risk % (no. of studies) |
|---|---|---|---|---|
| **Was the selection of stuttering & non-stuttering cohorts drawn from the same population?** | 4 | 8 | 1 | |
| **Is the sampling frame representative of the general population?** | 3 | | 10 | |
| **Can we be confident that those included in the ‘stuttering’ group had a diagnosable stutter?** | 12 | 1 | | |
| **Did the study match stuttering and non-stuttering participants for all variables that are associated with the outcome?** | 3 | 9 | 1 | |
| **Can we be confident in the assessment of anxiety or depression?** | 13 | | | |
| **Does the study report missing outcome data?** | 11 | 2 | | |
| **Are reports of the study free of suggested selective outcome reporting?** | 10 | 2 | 1 | |
| **Have the authors minimized potential bias in the statistical model selected to analyse study data?** | 8 | 4 | 1 | |
| **Other sources of bias?** | | 1 | 12 | |

*Note - data extracted into pre-prepared form that was modified from the template provided in Distiller-SR. Citation: Busse JW, Guyatt GH. Tool to Assess Risk of Bias in Cohort Studies. https://www.evidencepartners.com/resources/methodological-resources/*