

**Investigating risk for elevated anxiety and depression
symptoms in children who stutter relative to non-
stuttering peers**

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A Thesis submitted for the degree of
Doctor of Philosophy

Division of Psychology & Language Sciences
University College London (UCL)
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Declaration

I, Ria Frances Louisa Bernard, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Abstract

Speech, language and communication disorders are associated with greater risk for poor mental health outcomes. Adverse mental health can have a detrimental impact on educational and employment outcomes, and life chances. There has been much research examining the association between anxiety and developmental stuttering, which affects approximately 8% of children. Environmental factors commonly experienced by children who stutter, such as negative peer reactions, teasing and bullying, may put this clinical population at elevated risk of internalising problems. Yet, depression, which frequently co-occurs with anxiety and typically develops in late adolescence, has received comparatively little attention in relation to stuttering. Enhancing our understanding of the association between stuttering and symptoms of anxiety and/or depression, and the underlying mechanisms that may moderate any association, would inform timely, effective clinical management.

This thesis begins by systematically reviewing the extant literature. The second study utilises data obtained through an online questionnaire study to examine anxiety and depression symptoms in children who do and do not stutter, and analyse the association between child-, family- and contextual-factors and symptom scores in a sample of school-aged children who stutter. The final study draws on Millennium Cohort Study data to plot internalising symptom trajectories in cohort members who stutter, and considers the effect of co-occurring speech and language problems on development of internalising symptoms. The effect of multiple predictors on risk for internalising problems are then considered in a sample of adolescents who stutter.

Findings from this research indicated there was little evidence that stuttering affects internalising symptoms in a community sample. Female sex, maternal mental health, and co-occurring speech and language problems were associated with elevated internalising symptoms in children and adolescents

who stutter. Clinicians need to be alert to these factors in clinical assessment and ongoing management to promote mental well-being and resilience.

Impact Statement

The present research contributes to the existing body of literature examining the association between anxiety and stuttering in three ways. Firstly, it has complemented the work to date on the association between stuttering and anxiety by also investigating its association with depression, a body of research much less extensive by comparison. Secondly, the present research has examined factors that may serve to increase risk for internalising problems in childhood stuttering, thereby extending the existing research to consider the role of moderating factors. Thirdly, it provides further evidence from a community sample to enhance our knowledge of population estimates of internalising problems in childhood stuttering.

Theoretically, the current body of work has attempted to address some of the research gaps in the field. There is a substantial body of literature examining the association between stuttering and anxiety. However, much of this has relied on small, clinically-ascertained samples and has been characterised by a lack of longitudinal, community studies. The analysis of longitudinal population data and examination of factors that may serve to increase risk for internalising problems prompts a number of further research questions. These include consideration and further examination of the factors and underlying mechanisms involved in the association between mental health and stuttering in children and young people.

There are several clinical implications from this work. Primarily, speech and language therapists need to be alert to the factors that may serve to increase risk for internalising problems at the point of referral. This should include ensuring that promotion of positive mental health and wellbeing is embedded into clinical management. Particular attention should be given to referrals for girls who stutter, children with co-occurring speech and language problems, and adolescents with a family history of adverse mental health.

This research highlights the individuality of children who stutter and the ways in which factors intrinsic to, and independent of, stuttering may affect the

mental health and well-being of children who stutter. This is a particularly important message to convey to education professionals, both in terms of being alert to the variable impact stuttering may have on children, and also the ways in which they can create a supportive and inclusive learning environment.

Finally, this research contributes towards the evidence base around the social, emotional and mental health profiles of children who stutter by emphasising the importance of taking a holistic approach to assessment and management of this clinical population. Knowledge of risk and protective factors will help children and adolescents who stutter, families, educational and health professionals in facilitating positive mental health outcomes and building resilience. Nonetheless, future research is needed to enhance our understanding of risk and protective factors, and mechanisms involved in the association between internalising symptoms and stuttering.

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Dedication

For my Mum, Frances Rose Bernard.

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Key terms

- **Stutter.** ‘Stutter’ and ‘stammer’ refer to the same neurodevelopmental speech disorder. Although more commonly used in American English, throughout the thesis, ‘stutter’ will be used for consistency with the language used in the published studies.
- **Communication disorders.** This is an umbrella term that refers to disorders affecting speech, language and social communication.
- **Speech, language and communication needs (SLCN).** A broad term commonly used in the UK, and used here in reference to difficulties with one or several aspects of communication.
- **Speech sound disorders (SSD).** An umbrella term referring to communication disorders characterised by errors in speech production that affect a child’s intelligibility. There are a number of different types of SSD, but given the scope of the present thesis SSD is used in reference to two specific SSDs in particular:
 - **Phonological disorder.** A type of speech sound disorder concerning the production of sounds in words. The child can produce the sound but uses it incorrectly in a word, e.g. /k/ becomes /t/ so that ‘kite’ becomes ‘tite’. There are a number of typical speech sound errors (phonological processes) that children exhibit while they are learning to speak. In a phonological disorder, these common speech sound errors persist beyond the point at which most children have resolved them, or present with errors that are atypical at any age.
 - **Articulation disorder.** A type of speech sound disorder characterised by errors in the production of individual speech sounds. To produce sounds, children need to coordinate their tongue, lips, teeth, palate as well as the respiratory system. If a child cannot produce a particular sound they might substitute it, e.g. ‘wabbit’ instead of ‘rabbit’, or they may distort a sound so that it deviates from the expected production of that sound.
- **Clinical scores.** At some points in the thesis, reference is made to clinical (vs. subclinical) symptoms. Clinical scores refer to symptom

scores that exceed the clinical cut-off or threshold on that particular self- or parent-report measure. Sub-clinical scores are within the normative limits for the measure. The present thesis does not utilise diagnostic assessments or purport to evaluate rates of clinical anxiety or depressive disorder. Therefore, even in instances where symptom scores exceed the clinical cut-off, it should not be assumed that participants have a diagnosable anxiety or depressive disorder. Nevertheless, it is useful to distinguish scores that are elevated (but within normal limits) from scores indicative of clinical significance when evaluating the mental health and wellbeing of children who stutter and the general population to enhance our understanding of the specific psychosocial profiles of this clinical population.

- **Internalising disorders.** Internalising disorders are one type of emotional and behavioural disorder, which involve an individual 'internalising' problems or keeping issues to themselves. Internalising disorders predominantly refer to anxiety disorders and depression disorders as classified in the DSM-5. An internalising *disorder* is one that meets the symptom criteria set out in the DSM-5, and does not hold the same meaning as similar terms used in this thesis – namely internalising problems or symptoms.
- **Internalising problems vs symptoms.** In chapter 5, a distinction is made between internalising problems and internalising symptoms:
 - *Internalising symptoms* refers to symptom scores obtained on the referenced self- or parent-report measures, e.g. SDQ.
 - *Internalising problems* refers to scores that exceed the clinical cut-off for a particular self- or parent-report scale.

Chapter 1

General Introduction

This introductory chapter begins by defining developmental stuttering and considering it within the broader context of neurodevelopmental conditions, particularly communication disorders. This is followed by an overview of the research investigating the relationship between communication disorders and adverse mental health outcomes. The final section of this chapter outlines the hypotheses pertaining to any association between stuttering and adverse mental health outcomes in children and young people.

Defining developmental stuttering

Childhood stuttering is a neurodevelopmental condition characterised by disturbances in spoken fluency. Guitar (2014) defines stuttering in relation to three components: core behaviours, secondary behaviours, and feelings and attitudes, which illustrates the extent to which stuttering impacts communication beyond dysfluent speech. Core behaviours are involuntary speech behaviours and include *repetitions* of sounds and syllables; *prolongations* of sounds; and *blocks*, in which there is an abrupt stop to airflow or voice. Secondary behaviours, have been divided into *escape behaviours*, which Guitar (2014) argues occur in an effort to terminate the moment of stuttering, and *avoidance behaviours*, which reflect attempts to avoid the stuttering moments prior to it occurring. Secondary behaviours are characterised by physical concomitants, such as head nods, eye blinking, jaw tension, insertion of filler syllables ('uh') and word substitutions. Finally, negative feelings and attitudes represent the negative feelings of shame and frustration that can accompany stuttering, as well as negative self-perceptions and peer reactions (Guitar, 2014).

Recognising that stuttering can be characterised by more than disrupted verbal fluency in some young people prompts consideration of how it may affect mental health and wellbeing. Guitar's (2014) definition conveys the importance of understanding the wider implications of stuttering on social

and emotional development, as well as the motivation for researching potential risks for adverse mental health outcomes. An earlier and widely accepted definition offered by Wingate (1964) defines stuttering according to three parts (disruption in fluency; accessory activities; emotional state), which correspond to some extent to those detailed by Guitar (2014). However, while the speech behaviours detailed in the first part of the definition (disruptions in fluency) are considered core features experienced universally by those who stutter (Wingate, 1964), it has been suggested that accessory features are idiosyncratic (Wingate, 2002). This differentiation between universal and idiosyncratic characteristics of stuttering may contribute to the varying social, emotional and mental health outcomes observed in young people who stutter.

Stuttering prevalence and aetiology

It is estimated that around 8% of children will stutter at some point (Yairi & Ambrose, 2013), while stuttering prevalence is estimated to stand at around 1% (Bloodstein & Bernstein-Ratner, 2008). The onset of stuttering typically occurs between two and five years of age, with most onsets occurring around three years of age (Guitar, 2014; Yairi & Ambrose, 2005). The likelihood of beginning to stutter declines as children grow older (Bloodstein & Bernstein-Ratner, 2008), although later onset up to 13 years has been reported in some studies (Howell, 2011). The condition is more common amongst boys than girls; increasing from a reasonably equal ratio at onset to 4-5:1 males to females in adulthood (Yairi & Ambrose, 2005; Bernstein Ratner & Tetnowski, 2006). Consequently, it appears girls are more likely to recover.

There are numerous theories about the causes of stuttering that place varying emphasis on motor, linguistic, biological, psychological and emotional factors (see Andrews et al., 1983; Ambrose, 2004; Smith & Weber, 2017). Multifactor models advocate that stuttering occurs due to the interaction between these factors, with varying emphasis placed on different factors according to the particular model (Furnham & Davis, 2004). Recent research advances have led to the generally accepted view that stuttering

arises due to an interaction between neurological, genetic, environmental and psychological factors, which influence the development of stuttering (Ambrose, 2004). Additionally, social and emotional factors are considered to play a role in onset and maintenance (Furnham & Davis, 2004). However, the precise cause of stuttering remains a question for researchers.

The genetic basis of stuttering has been evidenced by twin, family aggregation and genetic linkage studies, and while a number of genes have been identified in the aetiology of the condition, research is on-going as to the involvement of additional genes and the mode of transmission (Frigerio-Domingues & Drayna, 2017; Domingues & Drayna, 2015). It is estimated that approximately 50% of people who stutter have a relative who stutters (Bloodstein & Bernstein-Ratner, 2008) and twin studies demonstrate that monozygotic twins display higher concordance of stuttering compared to dizygotic twins (Domingues & Drayna, 2015), with heritability estimates of 70% (Howell, 2011).

Recent research has shown neuroanatomical differences between stuttering and fluent individuals, characterised by differences in neural circuits affecting planning and execution of “self-initiated, intrinsically timed sound sequences” (Chang et al., 2019, pg.575). A review of neuroimaging research indicated structural and functional differences in brain organisation among adults and children who stutter, which appear to exist during speech as well as non-speech tasks (Etchell et al., 2018). Furthermore, research into neural connectivity in children who stutter indicate atypical connectivity in regions associated with fluent motor speech control, which has provided support for the view that stuttering is a complex neurodevelopmental disorder (Chang et al., 2018). Specifically, differences in development and integrity of white matter tracts, particularly in the left hemisphere, have been observed between children who stutter and fluent controls (Chang et al., 2015; Chang et al., 2008). Emerging research comparing groups of persistent and recovered stuttering children offer some insight into the different neuroanatomical mechanisms underlying persistence (Chang et al., 2019; Chow & Chang, 2017). Based on research to date, compared to fluent

children, children with a persistent stutter present with immature speech-motor coordination; reduced growth in white matter tracts involved in auditory-motor integration; and atypical cortical thickness in motor and pre-motor regions in the left hemisphere (Chang et al., 2019). In contrast, children who recovered from stuttering exhibited expected patterns of white matter development with age (Chang et al., 2019). These findings are not only important in terms of understanding the neural mechanisms underlying onset and risk of persistence, but also in relation to clinical management of stuttering as such findings could contribute to development of new therapy approaches.

Recovery and persistence of stuttering

Most natural recovery of stuttering takes place between a few months and three years' post-onset (Bernstein Ratner & Tetnowski, 2006; Yairi & Ambrose, 2005). However, studies have shown children may recover up until the teen years (Andrews & Harris, 1964; Howell et al., 2008). Around 70-80% of children will recover fluent speech following onset of stuttering (Bloodstein & Bernstein-Ratner, 2008), although such high estimates of recovery are associated with earlier onset (Yairi & Ambrose, 2005). In an effort to quantify the extent to which recovery rates decrease with age, Andrews et al. (1983) reviewed recovery rates across published studies and estimated that 75% of individuals who stutter at 4 years, 50% of those who stutter at 6 years, and 25% of individuals stuttering at 10 years old will have recovered by sixteen years of age.

While the majority of children who begin to stutter in childhood will resolve their stutter, a proportion of them will persist into adulthood. There have been a number of studies investigating the risk factors associated with the onset (Reilly et al., 2009) and persistence (see Walsh et al., 2018; Howell, 2011; Yairi et al., 1996) of stuttering. Factors that increase the risk of persistent stuttering include family history of persistent stuttering; male sex; later onset; failure to reduce frequency and severity of core speech behaviours over the first year; dysfluency continues for more than one year post-onset; duration of stuttering moments; and poor phonological skills (Guitar, 2014).

A risk factor approach for assessing the likelihood of stuttering persisting is helpful in clinical management, particularly in terms of informing triage and treatment approaches. Similarly, risk of adverse mental health outcomes may differ in persistent stuttering compared to individuals who recover fluent speech. Perhaps mental health outcomes improve when stuttering resolves, which is an important consideration given the high rates of recovery. Elevated anxiety levels and rates of anxiety disorder have been found in adults with a persistent stutter in many studies using a variety of measures (Howell, 2011; Craig & Tran, 2014; Iverach & Rapee, 2014).

Davis, Shisca and Howell (2007) compared trait and state anxiety in groups of children who stutter classified as 'persistent' or 'recovered', and found higher state anxiety scores amongst the persistent group while there were no differences between the recovered group and controls. State anxiety is a transitory state of arousal that occurs in specific situations (Endler & Parker, 1990). It was therefore suggested that elevated state anxiety in the persistent group suggests anxiety in particular situations is related to the stutter itself (Howell, 2011). However, data from the Illinois Longitudinal Study (Yairi and Ambrose, 2005) indicated no differences in anxiety scores between persistent, recovered and comparison groups on the Child Anxiety Scale at age four to six years, nor any differences on the Revised Children's Manifest Anxiety Scale (RCMAS) between the persistent and recovered groups at follow-up (aged 10 to 14 years). It should be acknowledged that the Yairi and Ambrose (2005) findings for the RCMAS are based on responses from 17 recovered and only five persistent participants, which limits the extent to which their findings can be generalised.

Consequently, if adverse mental health outcomes are associated with persistent stuttering then use of a risk factor approach may inform multi-disciplinary assessment, management and potential onward referral earlier in the diagnostic process.

Stuttering in the context of other neurodevelopmental conditions

Neurodevelopmental disorders comprise a cluster of conditions typically characterised by early childhood onset and impaired development of cognitive or motor function (Thapar & Rutter, 2015). As specified in the DSM-5 (APA, 2013), communication disorders are one such cluster of conditions. Communication disorders can be sub-divided into speech, language and social-communication disorders, of which stuttering is classified under speech disorders (Norbury & Paul, 2015).

According to data published by the Department for Education in 2019, speech, language and communication needs (SLCN) are the most common type of primary need¹ across children with special educational needs (SEN) in primary, secondary and special schools in England. It is estimated that almost 10% of five years olds present with a language disorder (Norbury et al., 2016). Population studies estimate prevalence of Developmental Language Disorder (DLD) to stand at around 7%; while approximately 2% have speech and language difficulties associated with another disorder, such as autism (Tomblin et al., 1997; Norbury et al., 2016). Based on findings from a recent population cohort, prevalence of speech sound disorder among 8 year olds is estimated to stand at 3.6% (Wren et al., 2016).

Speech, language and communication skills are essential to social and cognitive development, and predict later educational and occupational outcomes (Norbury & Paul, 2015). Consequently, communication disorders can have a detrimental impact on a child's lifetime opportunities and longer-term quality of life, which will be explored further in this chapter.

¹ Department for Education and ONS (2019): https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/814244/SEN_2019_Text.docx.pdf [accessed 11.6.20]

Co-morbidity across Neurodevelopmental Disorders

It is widely recognised that there is substantial comorbidity between neurodevelopmental disorders (Thapar & Rutter, 2015). Moreover, research has indicated that a proportion of children who stutter present with co-occurring neurodevelopmental disorders, particularly other communication disorders (Briley & Ellis, 2018; Blood et al., 2003a). However, some researchers have argued that there is insufficient evidence of an association between high rates of language disorder among children who stutter, and have shown that many such children present with advanced language abilities (Nippold, 2019; Watkins, 2005).

Much of the earlier research into co-existing communication disorders in children who stutter is based on survey data collected from clinical samples. For example, Arndt and Healey (2001) found that 44% of 467 children who stutter presented with an additional phonological and/or language disorder, based on survey responses from speech and language therapists (SLT) across 10 US states.

In a larger USA-wide study of 2,628 children who stutter, a higher estimate of 62.8% of children presented with at least one co-occurring speech, language or non-speech-language disorder (Blood et al., 2003a). Moreover, a significantly higher proportion of males than females who stutter presented with co-occurring speech and/or language disorders, but this significant sex difference was not observed for co-occurring language disorders (Blood et al., 2003a). The most common co-occurring speech disorder alongside stuttering was found to be articulation disorder (33.5%), and the most common co-occurring language disorders were expressive semantic (13.5%) and receptive semantic (12.1%) disorders. A further 34.5% presented with another condition, such as learning disability and attention deficit disorder. Co-occurrence with some neurodevelopmental disorders, including autism, Tourette's syndrome and epilepsy, were reported with less than 1% frequency. The validity of these findings was improved by the fact that information relating to existence of co-occurring disorders needed to be

properly documented by clinicians rather than self-reported by caregivers. However, surveys were only sent to clinicians employed by schools as opposed to other clinical settings. This increases selection bias, as children presenting with more than one disorder are likely to present with greater, and potentially more complex, difficulties in school and thus warrant attention from school-based speech and language therapists. Additionally, estimated rates of co-occurring disorders were not compared to a control group of non-stuttering peers, although the authors made some comparison with population norms. Some researchers have argued that rates of phonological and language disorders in this population may be overestimated, particularly when findings are based on caseload surveys, as children presenting with more than one co-occurring condition are more likely to be referred for treatment (Nippold, 2004).

A more recent population study compared data on presence of developmental disorders for children who stutter and children who do not stutter obtained from the National Health Interview Survey (NHIS) over six years (Briley & Ellis, 2018). These findings indicated that children who stutter were more likely to present with one developmental condition (52%) than children who do not stutter (15%). A total of 29% of children who stutter presented with at least two developmental conditions, compared to 6% of fluent children. Briley and Ellis (2018) concluded that the odds of having at least one developmental disorder was 5.5 times higher in children who stutter compared to non-stuttering peers. These findings were based on data from 1,231 children who stutter, which is smaller than the Blood et al. (2003) study. Furthermore, unlike the previous studies, stuttering was confirmed through self-report rather than clinical diagnosis by an SLT, which may affect the validity of the 'stuttering' sample.

The relationship between speech sound difficulties and stuttering has been extensively examined, with poor phonological skills and lower speech sound accuracy identified as significant predictors for persistent stuttering (Singer et al., 2022; Singer et al., 2020; Guitar, 2014). Early reviews of the literature suggested that children who stutter are more likely to present with speech

sound disorders relative to children who do not stutter (Louko, 1995; Wolk, Conture & Edwards, 1990). Yet estimated rates of co-occurrence vary widely, which has been attributed to differences in the definition of SSD used in studies, methods for eliciting speech samples, and differences in comorbidity rates within clinical versus community samples (see Nippold, 2001). A recent examination of a community cohort found 6.88% of children diagnosed with a stutter between two and four years had a co-occurring SSD (Unicomb et al., 2020), which is much lower than the 30% - 35% estimated in clinical samples (Yairi & Ambrose, 2005).

Overall, research suggests that a proportion of children who stutter present with a co-occurring neurodevelopmental disorder, which may increase the impact on social, emotional and mental health development. The observed rates of co-morbidity between stuttering and other communication disorders is noteworthy given that children with speech, language and communication disorders present with increased social, emotional and behavioural problems compared to peers (Norbury et al., 2016; Levickis et al., 2018). Therefore, this would suggest that any association between stuttering and adverse mental health outcomes needs to consider the role of co-occurring conditions.

Communication disorders & mental health outcomes

The literature suggests there is an association between communication disorders and later adverse mental health outcomes (Toppelberg & Shapiro, 2000). Compared with typically-developing peers, higher rates of anxiety and/or depression have been reported amongst children and young people with language disorder (Clegg et al., 2005; Conti-Ramsden & Botting, 2008; Botting et al., 2016), poor early receptive language skills (Schoon et al., 2010), and high-functioning autism and Asperger's syndrome (Kim et al., 2000; Lugnegård et al., 2011). Equally, there is a high rate of language disorder, which often goes undiagnosed, among children referred to tertiary child and adolescent mental health services (Cohen et al., 1993).

This section discusses the existing literature examining the association between adverse mental health and two specific communication disorders – speech sound disorder and language disorder.

Mental health outcome trajectories in childhood language disorders

Longitudinal designs have provided insight into longer-term mental health outcomes associated with childhood-onset communication disorders. Several longitudinal studies have indicated that a history of communication disorder is associated with elevated rates of DSM-diagnoses, especially anxiety and depression, in adolescence and adulthood.

Data from the Manchester Language Study reported that young people who had a history of Specific Language Impairment (SLI; also known as Developmental Language Disorder) displayed significantly higher symptom levels of anxiety and depression at 16 years of age compared with peers matched for age and sex (Conti-Ramsden & Botting, 2008). Furthermore, a larger proportion of the SLI group presented with scores above clinical threshold on measures of anxiety and depression. In a later study using data from the same sample aged 24 years, depression and anxiety scores were found to be significantly higher in adults with language impairment compared to adults with typical language function (Botting et al., 2016). Additionally, they found that significantly more adults in the language impairment group presented with clinical levels of depression relative to controls.

The Ottawa Language Study assessed speech and language skills in a community sample of five-year old children, generating a sample of children with communication disorders and a sample with typical language development (Beitchman et al., 1986). This longitudinal study re-tested participants at seven year intervals, reporting mental health outcomes between five and 31 years of age, according to the DSM-III-R (APA, 1987) psychiatric disorder classifications. Data indicated elevated rates of psychiatric disorders amongst participants with language disorder at ages

five, 12 and 19 years of age compared with controls (Beitchman et al., 1986; Beitchman et al., 1996; Beitchman et al., 2001). Furthermore, data from the seven year follow-up indicated that those with speech/language impairments at age five were more likely to be diagnosed with a psychiatric condition at age 12.5 years (42.9%) than the comparison group (20.7%), even if their speech/language skills had improved (Beitchman et al., 1996). Specifically, the children who had speech/language difficulties at age five, were more likely to have emotional diagnoses (anxiety or depression) at age 12.5 years than children assessed to have typical language development at five years.

Young people with a history of early language impairment may be at increased risk for social phobia in particular (Voci et al., 2006). Adolescents classified as language-impaired at age five were more likely to meet criteria for Social Phobia at age 19 years (16%) than the comparison group (6.5%). Severity of language impairment was not related to prevalence of social phobia. However, those with social phobia, regardless of language group, were significantly more likely to meet criteria for another psychiatric disorder, the most common of which was major depressive disorder (Voci et al., 2006). Yet, by age 31 years, Beitchman et al. (2014) found similar rates of psychiatric disorders between individuals with a history of language disorder and controls. This is in contrast with other studies, which found that higher rates of anxiety and depression persisted into adulthood amongst those with a history of language disorder (Clegg et al., 2005; Botting et al., 2016). The authors put forward several possible reasons for this trend. Firstly, the analyses may have had insufficient power to detect small effects, which perhaps did exist between the language-disorder and comparison groups. Secondly, emotional maturation and development of compensatory strategies may have mitigated the impact of language disorder, while moving out of formal education may have reduced the impact of communication pressures associated with social and educational demands in school.

Finally, the severity of language impairment may have contributed to these contrary outcomes relative to the existing literature. The present cohort represented predominantly mild-moderate language disorder and

consequently more severe forms of language disorder may have affected psychiatric outcomes. Furthermore, while the Ottawa Language Study involved a community sample and focused specifically on mental health outcomes among participants with speech and/or language disorders, both Clegg et al. (2005) and Botting et al. (2016) recruited children from special schools and units and included pragmatic disorders in the sample.

In light of these findings, it should be acknowledged that absence of a psychiatric disorder does not necessarily equate to absence of subclinical symptomology. Investigation of the association between language disorder and social anxiety at age 19, 25 and 31 years indicated that the elevated rate of diagnosed social phobia in the language disorder cohort was not apparent after 19 years of age (Brownlie, Bao & Beitchman, 2016). However, subthreshold social anxiety symptoms did persist beyond 19 years of age in this cohort, with the authors concluding that young adults with a history of language disorder are likely to continue to experience symptoms of social anxiety but such symptoms may not be sufficiently severe to meet diagnostic criteria (Brownlie et al., 2016).

A systematic review and meta-analysis evaluated the longitudinal evidence pertaining to psychological outcomes associated with early language impairment, comprising 19 follow-up studies reporting outcomes for eight cohorts (Yew & O'Kearney, 2013). The findings indicated that children with SLI were twice as likely to meet criteria for internalising problems compared with typically developing peers. Moreover, children with a history of SLI more frequently displayed clinical levels of psychological problems during childhood and adolescence. However, findings did not elucidate the risk of specific mental health disorders. Interestingly, a recent longitudinal community study concluded that children with persistent language disorder from preschool to early primary school were more likely to present with social, emotional and behavioural problems, however emotional symptom scores did not differ between those with and without language disorder (Levickis et al., 2018).

In summary, current evidence suggests there is not only an association between existing communication disorders and adverse mental health, but that a history of speech and language disorders in early childhood affects the likelihood of developing psychiatric symptoms in later life. In relation to stuttering, it may be that even children who recover from stuttering may experience some level of adverse mental health due to childhood experience of dysfluency. Furthermore, similarly to other communication disorders, social anxiety may be the predominant issue in children and adolescents who stutter, especially considering the elevated rates of social anxiety disorder observed in adults who stutter (e.g. Iverach & Rapee, 2014).

Mental health outcome trajectories in childhood speech disorders

Prior research has indicated that the risk of psychiatric disorders and poorer mental health outcomes is higher in individuals with a history of language disorder as opposed to speech disorders (Baker & Cantwell, 1982). While almost half of the clinical sample met criteria for a psychiatric disorder, prevalence differed by category of communication disorder. Baker and Cantwell (1982) reported that 95% of those with a pure language disorder had a diagnosable DSM-III (APA, 1980) psychiatric disorder compared with 45% of those with a speech *and* language difficulty and 29% of those individuals with only a speech disorder. In their review of the literature, Toppelberg and Shapiro (2000) concluded that disorders of grammar, semantics and pragmatics, but not phonology, are significantly associated with childhood psychiatric disorders.

These differences in psychiatric disorder prevalence amongst those with speech, as opposed to language disorders, were also observed in the Ottawa Language Study. Beitchman et al. (1996) reported that of the children with a speech/language impairment at age 12.5 years, children categorised as having both speech *and* language disorders were most likely to have a psychiatric disorder (57.1%); followed by participants with only a language disorder (42.1%); and those children presenting with *only* a speech disorder (25.9%). Within this speech disorder sub-group, the researchers examined whether the type of speech disorder (voice vs. fluency vs. articulation

disorder) at age 12.5 years was associated with psychiatric outcomes, and found no association. Moreover, no relationship was found between specific speech problems diagnosed at age five years and psychiatric outcomes at age 12.5 years (Beitchman et al., 1996). These findings provide further evidence that higher prevalence of psychiatric disorders exists amongst childhood language disorders compared to pure speech disorders.

These observed differences in psychiatric outcomes amongst participants with language disorder compared with speech disorder continued to be reported at 19 years of age (Beitchman et al., 2001). Logistic regression analyses indicated that language impairment at age five years was associated with significantly higher rates of anxiety disorder relative to controls. However, this finding was not replicated with the speech-impaired-only group; no differences were observed between the speech-impaired and comparison groups. The speech-impaired group comprised children with speech disorders, including stuttering, voice disorders and dysarthria, in the absence of additional language difficulty. While this would suggest reduced risk of anxiety in individuals with a history of speech disorders (including stuttering) relative to language disorders, the language-disordered group comprised 77 children who had either presented with language disorder only, or with both speech *and* language disorders. It should also be recognised that of the 244 datasets collected at age 19 years, only 38 participants had been diagnosed with only a speech impairment at five years, of which the number who stutter is unknown. Therefore, the small sample size makes it difficult to draw robust conclusions about the risk associated with speech disorders and stuttering in particular. Nevertheless, perhaps in cases where stuttering co-occurs with another communication disorder the risk of anxiety is increased as a result of this co-morbidity, rather than stuttering per se.

More recently, Lewis et al. (2016) compared psychosocial outcomes in adolescents and adults with a history of speech sound disorder (SSD) and those with SSD and language impairment (LI). They found that early history of LI was associated with a greater risk of psychosocial issues than with SSD-only or controls. This led them to conclude that poor psychosocial

outcomes in adolescents with SSD were related to comorbid LI rather than SSD itself. However, in contrast with other studies the findings were based on rating scales rather than diagnostic interviews and so do not provide information on psychiatric diagnosis rates. Nevertheless, the finding that adults and adolescents with SSD-only present with good psychosocial outcomes suggest that co-occurring language impairment in individuals with speech disorders may increase the risk of adverse mental health outcomes in young adults.

While acknowledging that stuttering is one of many communication disorders, the elevated risk of poor mental health outcomes associated with a wide range of neurodevelopmental conditions provides rationale for further investigation into the mental health profiles of children who stutter. The lower rates of mental health conditions amongst children presenting only with speech sound disorders may, on the other hand, indicate that children who stutter are not necessarily at heightened risk of adverse mental health issues. Instead, it may be that co-morbidity between stuttering and other neurodevelopmental conditions, such as language disorder, moderates this risk.

Mechanisms underlying the association: communication difficulties and mental health outcomes

Durkin & Conti-Ramsden (2010) argue that the complex nature and heterogeneity of emotional and psychiatric outcomes in young people with specific language impairment (SLI) can be attributed to the interaction between social and emotional functioning and developmental and environmental factors in adolescence. Therefore, the extent to which children and adolescents with communication disorders experience adverse mental health outcomes is likely to differ by individual child depending on their profile of language needs, environmental, social and genetic factors. Botting et al. (2016) suggested two possible mechanisms that underlie the link between emotional health and language abilities: (i) gene-environment influences, such as parental mental health, difficulties with peer relations and developing

friendships; and (ii) internal child factors, including difficulty with emotional regulation and understanding, resilience and presence of co-morbid difficulties.

The Manchester Language Study indicated that adolescents with SLI were at elevated risk of anxiety and depression symptoms and more likely to score above clinical threshold relative to typically-developing young people. However, language ability was not identified as a direct predictor of mental health outcomes (Conti-Ramsden, 2008). This would suggest that SLI per se may not increase the risk of anxiety/depression, but instead other factors contribute to heightened vulnerability amongst some individuals with SLI, such as family history of mental health issues and bullying (Conti-Ramsden, 2008).

Snowling et al. (2006) found that persistence and severity of the initial speech-language delay is associated with later psychosocial outcomes; in cases where speech and language difficulties resolved by 5.5 years children presented with good outcomes at age 15 years. These findings indicated that risk of psychiatric problems in adolescence was associated with persistent and severe language difficulties and low nonverbal IQ as opposed to pre-school history of communication difficulties. However, other studies have not found an association between severity of speech/language impairment and later psychosocial outcomes (Conti-Ramsden, 2008; Voci et al., 2006).

Childhood bullying is associated with poorer mental health outcomes (Copeland et al., 2013) and children with DLD may be more likely to be bullied (Conti-Ramsden & Botting, 2004). Recently, Kilpatrick et al. (2019) failed to show that a history of DLD was directly associated with internalising or externalising symptoms; instead finding a significant interaction between history of DLD and bullying victimisation for internalising symptoms. This would suggest that where bullying victimisation occurs it may be associated with increased risk of internalising symptoms in young people with a history of DLD.

Botting et al. (2016) found that not only did adults with language impairment present with higher scores on anxiety and depression measures, they also reported lower self-efficacy than typical controls. Moreover, they found that the relationship between language ability and emotional health was mediated by self-efficacy. Given that anxiety and depression levels were lower amongst individuals with higher self-efficacy, it would suggest that self-efficacy may play a protective role in development of psychiatric problems in this population (Botting et al., 2016).

In summary, research has suggested that while language disorders are associated with increased risk of poorer mental health outcomes, and particularly elevated anxiety and depression, there may be additional factors that play a role in this relationship. Furthermore, the association between stuttering and mental health outcomes may be mediated by the interaction between a variety of factors, such as family history of mental health, bullying victimisation, timely intervention, and co-occurring disorders. These may act as risk and protective factors in the development and trajectory of mental health issues. The concept of risk factor modelling in stuttering, and in relation to mental health outcomes, will be discussed further in chapters four and five.

Stuttering and mental health outcomes

There has been a substantial amount of research investigating the association between stuttering and adverse mental health outcomes, especially amongst adults with a persistent stutter. Theories differ with respect to the direction of the relationship between stuttering and anxiety; specifically, whether anxiety is a cause (Sheehan, 1970); consequence (Perkins, 1979; Ryan, 1974); or plays a mediating role (Brutten & Shoemaker, 1967) in the development and persistence of stuttering.

It is important to consider the present research in the context of current definitions of stuttering, and the extent to which anxiety features in the diagnostic criteria. The updated DSM-5 (APA, 2013) classifies Childhood-

Onset Fluency Disorder (Stuttering) as “disturbances in the normal fluency and time patterning of speech that are inappropriate for the individual’s age and language skills” (p45) and lists a number of behaviours characteristic of the disorder. Crucially, in the context of this thesis, the DSM-5 has added that stuttering “causes anxiety about speaking...” (p46) to its definition. Given conflicting theories about the nature of any relationship between anxiety and stuttering, it does not appear that research has conclusively found stuttering to be a cause of anxiety. Moreover, studies have generated inconsistent findings regarding rates of anxiety and depression in people who stutter, especially children, relative to fluent peers, which will be discussed in chapter two.

Why might children who stutter be at risk of adverse mental health?

Stuttering falls under the broader umbrella of speech, language and communication disorders, which are associated with elevated risk of adverse mental health outcomes in children and young people. But to what extent, if at all, might stuttering specifically be associated with elevated rates of poor mental health? Perhaps the clinical features that characterise stuttering put this group at particular risk. Spoken communication is pivotal to society and therefore difficulties with verbal fluency can create challenges to full participation. As well as disturbances in verbal fluency, stuttering moments may be accompanied by overt physical concomitants, such as head nodding, jaw tension and eye blinking, which may affect an individual’s self-perceptions as well as how they are perceived, and responded to, by others.

The *International Classification of Functioning, Disability, and Health* (ICF; WHO, 2001) provides a framework for understanding the variable contextual factors that may affect the experiences of the individual who stutters (Yaruss & Quesal, 2004). By applying the ICF to stuttering, Yaruss and Quesal (2004) describe the experiences of this clinical population in relation to impairment in body function (e.g. core speech behaviours) and structure (e.g. brain structure), as well as subsequent limitations in activity and participation,

such as forming relationships, school and work engagement, and social interaction (Yaruss & Quasel, 2004). Moreover, this model details many of the personal and environmental factors that contribute to an individual's experience of dysfluency and the extent to which within- and between-speaker variability exists. The following section summarises some of the possible reasons individuals who stutter may be at risk of poor mental health outcomes.

Societal and peer reactions

Dysfluency may result in negative reactions from non-stuttering communication partners and assumptions about communicative and intellectual competence. Adults who stutter report anticipation and experience of public stigma, both of which have been found to be predictors of global mental health in this population (Boyle, 2018). Research indicates that people who stutter show increased fear of negative evaluation (Messenger et al., 2004), heightened communication apprehension and poor self-perceptions of communication competence (Erickson & Block, 2013; Blood et al., 2001). Adults who stutter have shown reduced self-efficacy for speaking, which has been attributed to embarrassment associated with dysfluency (Bray et al., 2003). However, other studies have found that people who stutter do not present with poorer self-esteem compared to non-stuttering peers (Blood et al., 2007).

It has been well-documented that children who stutter are more likely to be bullied (Blood & Blood, 2007; Cook & Howell, 2014) and rejected by peers (Davis et al., 2002). Peer rejection and bullying can lead to social isolation, which has damaging consequences for the child's social and emotional well-being. Bullying in this population may affect the development of peer relationships, self-esteem and depression in the short- and long-term (Hugh-Jones & Smith, 1999). Furthermore, similarly to the general population, research suggests that childhood victimisation has longer-term psychosocial consequences that persist into adulthood in people who stutter (Blood & Blood, 2016).

Educational and occupational outcomes

Given the social and emotional issues that may accompany peer victimisation, rejection, and negative self-perceptions in school, as well as stigmatisation, stuttering may also have implications for educational and occupational outcomes. For instance, children who stutter may not reach their potential in subjects that put more emphasis on spoken language skills, which may be reflected in lower attainment in speaking and listening targets. A recent study investigating educational attainment in higher education in the USA identified stuttering as a predictor of lower chances of college enrolment (Rosenbaum, 2018). Furthermore, stuttering was associated with decreased likelihood of obtaining a Bachelor's (BA) degree or above but not with achieving sub-BA credentials (Rosenbaum, 2018). The author attributed this to stigma around stuttering and the academic nature of BA-level courses, in which it is likely students would be expected to engage in more class discussion and request help, which may create barriers to students who stutter. Research has also indicated that self-reported stuttering severity is negatively correlated with educational attainment (O'Brian et al., 2011).

However, findings from a population study failed to show a significant effect of stuttering on educational outcomes (McAllister et al., 2012). In this study, the authors examined the impact of stuttering alongside known predictors of educational outcomes on school leaving age and highest qualification at age 50 years. They found that people reported to stutter at age 16 years were not significantly more likely to leave school at the earliest opportunity. Instead, other predictors of educational performance were found to be more powerful, including social class, parental education, economic circumstances and reading comprehension at age 11 years (McAllister et al., 2012). Therefore, the risk of lower educational outcomes in this population may reflect additional risk factors known to impact educational outcomes rather than stuttering itself.

Findings have been mixed with regards to the association between socio-economic status (SES) and childhood speech disorders, although more recent community studies have found an association between low SES and childhood SSDs (see Wren et al., 2016; Eadie et al., 2015; Keating, Turrell & Ozanne, 2001). There is insufficient evidence that SES is associated with stuttering; population studies found no significant between-group differences in SES between children who do and do not stutter (Andrews & Harris, 1964), while Howell (2007) reported that the occupation of the primary earner was not associated with persistence or recovery. However, socio-economic status may impact the extent to which children receive the support they need as families from deprived backgrounds may find it more difficult to access appointments due to inflexible working patterns. Higher maternal education level has been associated with risk of stuttering onset (Reilly et al., 2009) and stuttering severity (Richels et al., 2013). Although it has been suggested that the association between higher maternal education and stuttering onset reflects the fact that highly educated mothers may be more likely to identify and report stuttering symptoms (Reilly et al., 2009).

Adults who stutter have reported that they see stuttering as a barrier to employment opportunities and job performance (Klein & Hood, 2004). In a survey of 232 adults who stutter, 70% believed stuttering affected their chance of being hired and promoted, while 69% believed stuttering interfered with their job performance. However, within those trends, it was found that women, Caucasian participants and those with a mild stutter were less likely to perceive their stutter as having a negative impact in the workplace relative to men, non-Caucasian participants and those with a moderate/severe stutter (Klein & Hood, 2004). However, this sample was drawn from people who had at least minimal contact with a national support group for stuttering, and comprised people who were considered to have better educational attainment and higher status jobs than a random sample of the population (Klein & Hood, 2004). Consequently, the participants may have been more likely to perceive their stutter as a barrier due to the high pressures associated with higher-level jobs.

While these findings provide insight into the self-perceived impact of stuttering on employment outcomes, studies have also investigated the relationship between stuttering and employment status and progression. Stuttering has been associated with holding lower-status jobs at age 50 in the UK (McAllister et al., 2012) and an earnings gap in the US (Gerlach et al., 2018). However, despite the significant association between occupation status and stuttering, McAllister et al. (2012) found those who stuttered at 16 years of age were not significantly more likely than controls to experience unemployment lasting more than one month at the beginning of their career; earn significantly less at 23 or 50 years of age; or hold a job of lower status at 23 years of age.

Therefore, it may be that the beliefs that people who stutter hold about their own capability and assumptions about how they are perceived by others affects the extent to which they seek out higher-status jobs. After all, adults who stutter have reported that having a stutter affected the extent to which they sought employment requiring limited spoken communication and the decision to take a job or promotion (Klein & Hood, 2004). This may support the idea that stuttering is associated with increased anxiety due to apprehension about public responses and sufficient resilience to progress.

Early childhood experiences

Experiences in early childhood shape development and these experiences influence the risk of developing adverse mental health conditions (Mrazek & Haggerty, 1994). Negative peer reactions and bullying at an early age may have profound short- and long-term effects on a child's mental health, especially anxiety and depression levels (Shoeler et al., 2018; Lereya et al., 2015). Given that rates of bullying are estimated to be high amongst children who stutter, bullying may be a contributing factor to the relationship between stuttering and mental health.

Research based on population data from the Strengths and Difficulties Questionnaire (SDQ) suggests children who stutter are more likely to present

with behavioural, emotional and social well-being issues than children who do not stutter (Briley et al., 2019; McAllister, 2016). These differences in social, emotional and behavioural development may be apparent from as early as three years old (McAllister, 2016), and are more pronounced in children who stutter with co-existing developmental conditions (Briley, et al., 2019). This would suggest there is a need to address social and emotional issues early in development to help offset later adverse mental health outcomes. Moreover, early behaviour problems may influence responses from adults and peers and thereby interaction with the child, which could negatively affect social and emotional development.

Summary

Stuttering is a highly heterogeneous condition in terms of severity, persistence, symptomology and impact on quality of life. Consequently, differences in early childhood experiences among children who stutter and also between those who do and do not stutter may moderate the risk of anxiety and depression in the stuttering population, as illustrated in Figure 1.

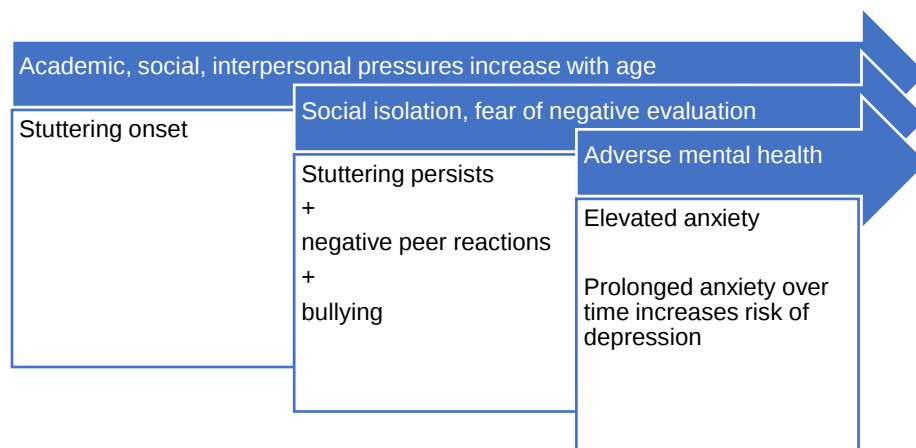


Figure 1: hypothetical association between anxiety/depression and stuttering across childhood - showing development of anxiety/depression symptoms moderated by increasing age and early childhood experience.

On the other hand, when considering early childhood experiences, it is important to recognise the role of protective factors in helping to offset mental health problems, as exemplified in Figure 2.

Self-efficacy is the belief that one has in their capability to execute a task or behaviour to achieve an outcome; it reflects an individual's self-confidence in

their ability to cope with stressors (Schwarzer & Warner, 2013; Bandura, 1977). Self-efficacy influences the amount of effort and persistence an individual has in the face of an obstacle or adverse experience (Bandura, 1978). Resilience refers to an individual's ability to cope with stressors and resist adversity, and thus it differs from self-efficacy in so far as resilience is not necessary in the absence of a stressor (Schwarzer & Warner, 2013). Since self-efficacy influences how an individual thinks, feels and behaves, it plays a role in overcoming stressful situations, thus helping an individual to be more resilient in the face of adversity (Schwarzer & Warner, 2013).

It has been suggested that self-efficacy mediates the relationship between emotional health and language ability in adolescents with a history of language impairment, with lower levels of anxiety and depression found amongst those with higher levels of self-efficacy (Botting et al., 2016). Craig, Blumgart and Tran (2011) compared two groups of adults with a chronic stutter; classified as 'resilient' or 'non-resilient' as determined by their global psychopathology score. High self-efficacy, high levels of social support, and superior social functioning were identified as factors that contribute to lower levels of psychopathology in this population (Craig, Blumgart & Tran, 2011). The interaction of risk and protective factors in any association between stuttering and mental health outcomes will be a focus for the current thesis.

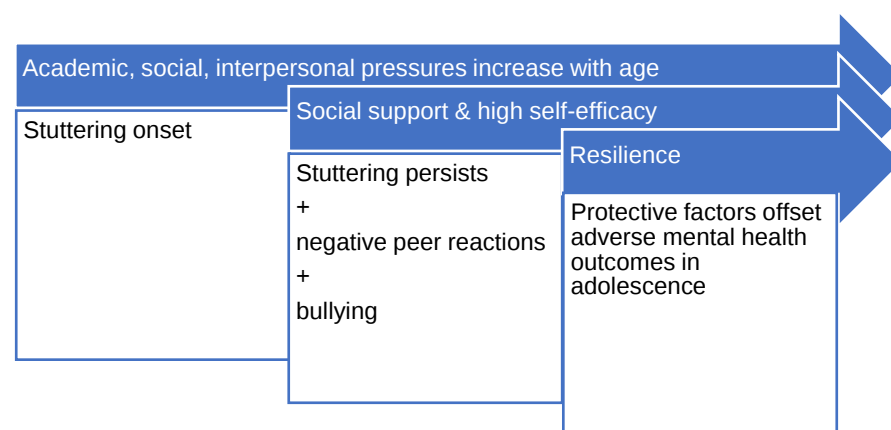


Figure 2: hypothetical association between anxiety/depression symptoms and stuttering across childhood - showing how development of anxiety/depression is offset by protective factors.

Thesis objectives

My PhD thesis aims to address the question of whether children who stutter (aged 8 – 13 years) are at elevated risk of anxiety and depression relative to non-stuttering peers with and without other speech/language disorders.

The two main objectives of the present PhD research include:

1. To synthesise the extant literature concerning whether children and young people who stutter are at greater risk of anxiety and depression than non-stuttering children with and without speech and language difficulties.
2. To consider additional factors that are associated with elevated symptoms of anxiety and depression that can be used to inform clinical management and multi-disciplinary referral when working with this clinical group.

In order to meet these objectives, this thesis aims to address the following research questions:

- Are children and adolescents who stutter more likely to experience anxiety and/or depression than non-stuttering peers (those with typical language and those with other language/speech disorders)?
- What additional variables moderate the relationship between stuttering and anxiety and depression?

The thesis is structured around three studies undertaken to address these research questions. The subsequent chapter comprises a systematic review and meta-analysis of published studies comparing symptoms of anxiety and depression in children and adolescents who stutter with fluent peers.

Chapters three and four detail the findings of an online questionnaire study investigating symptoms of anxiety and depression in a sample of children who stutter living in the UK. The fifth chapter comprises the results of secondary data analysis. Data from the Millennium Cohort Study were analysed, and internalising symptom trajectories were plotted for cohort members (3 – 17 years) reported to stutter in the UK. Finally, multiple risk

models evaluate the extent to which putative risk factors for adverse mental health are associated with internalising problems in adolescents who stutter. It is hoped that this will aid clinicians in management planning at the point of referral by identifying child and family factors associated with increased risk for internalising problems.

Impact of COVID-19 on research questions and data collection

The COVID-19 pandemic has had a substantial impact on the original plans for the present thesis, especially regarding data collection, study protocols and proposed analyses. In particular, the broader implications of this public health crisis negatively affected recruitment efforts due to uncertainty, disrupted routines and financial pressures for many families across the UK. This meant that some proposed analyses needed to be re-considered in an effort to obtain sufficient power, which is discussed in relation to the planned individual studies in each chapter. Furthermore, in-person testing was not feasible throughout much of the pandemic, and therefore secondary data has been analysed in an attempt to address the original research questions.

Critically, the empirical data were likely confounded by the impact of COVID-19 on mental health in the general population (Chapter 3 & 4). Research has indicated that compared to pre-pandemic levels, there was an increase in prevalence of mental health symptoms in adults in the UK (Pierce et al., 2020). Early studies including children and adolescents have pointed to potential increases in internalising symptoms during the pandemic (Bignardi et al., 2020; Newlove-Degado et al., 2021), although further longitudinal studies are underway to investigate the short- and longer-term mental health implications.

The initial national lockdown was announced by the UK Government in March 2020, and data from the stuttering sample was primarily collected via an online questionnaire over the entirety of the pandemic. The questionnaire data from study two was gathered from February 2020 onwards, at a time when the existence of the virus was internationally acknowledged but

restrictions had not been implemented. It remained open during the ensuing periods of national and regional lockdowns, school closures, and social distancing measures.

It is likely that mental health, well-being and levels of resilience will have fluctuated, rather than remained static, over the course of the pandemic, for example during periods of national or regional lockdown compared with easing of restrictions by the UK Government. Eligibility criteria for all studies required participants to reside in the UK, which reduced international disparities regarding the impact of the pandemic. Nevertheless, regional discrepancies in infection and mortality rates, local pressures on health providers and school closures need to be recognised.

Educational impact of COVID-19 for children and adolescents

Children and adolescents in particular experienced numerous disruptions to their education through school closures and social distancing rules in the classroom, which are likely to have adversely affected opportunities for social interaction and development and exacerbated the effect of social inequality on educational attainment. The extent of the impact of school disruption will differ across the general population, for example in relation to home environment and family socio-economic context. Increased reliance on online teaching, for example, may have affected the likelihood of actively participating in classroom activities as children who stutter may be negatively affected by seeing themselves stutter in real time on video, criticism from teaching staff who are unable to identify more covert aspects of stuttering and silent blocking, and fear of negative evaluation from peers. However, it is also reasonable to hypothesise that home-schooling may have had some positive influences on mental health and well-being for some children who may be at risk of negative experiences in the school environment. Children who stutter, who as a group have been found to be at increased risk of bullying and negative peer reactions, may have faced less social stigmatisation due to not being in the classroom thereby improving education experience.

Potential factors contributing to risk and resilience during the pandemic

It has been recognised in the literature that a variety of factors may be involved in vulnerability to mental distress and changes in mental health amongst children and adolescents in the context of COVID-19 (see Singh et al., 2020; Magson et al., 2020).

Numerous variables may potentially have influenced the mental health and well-being of individual children, regardless of stuttering status, during the pandemic. For instance, children whose parents were key workers faced additional stress and worry given the elevated risk to their parent's health, while the extent to which families were able to maintain access to their support networks may have affected parental mental health. Equally, children of parents who were considered to be most at risk from the virus likely faced the added burden of acute stress over transmission and fears around parental mortality. The high infection and mortality rates associated with this virus in the UK also meant that many families were affected directly by illness, bereavement and longer-term health consequences. Pressures on mental health services likely increased during the pandemic and, coupled with the inability of professionals outside the home to directly observe the behaviour and well-being of children and adolescents, insufficient support would have been available (Newlove-Degado et al., 2021).

Financial pressures brought about by unemployment was partly ameliorated by the furlough scheme. Nonetheless, this meant families were taking home 80% of their pre-pandemic income, substantially affecting low income households. As a consequence of home working and schooling many families may have spent the majority of time in poor-quality, cramped housing conditions, which may have adverse psychological and health consequences. Equally, stresses on the family dynamic brought about by relative isolation in the home are likely to significantly exacerbate existing issues. Reports of family violence increased internationally during the

pandemic (Usher et al., 2020). Isolation within the home due to lockdown also had implications for children at risk of emotional and physical abuse, as they were less likely to be identified by outside agencies.

In summary, aside from the physical health risks posed by COVID-19, the pandemic is likely to have had a multifactorial impact on children and adolescents due to a variety of biological, environmental and social factors (de Figueiredo et al., 2021). Due to the timing of data collection for the second study reported in this thesis, data could not discern the effect of individual differences in experience of the virus, which needs to be acknowledged when interpreting the findings. However, efforts have been made to counteract this confound by comparing group data collected only during the pandemic period and modifications to the planned analyses.

Chapter 2

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

Citation: Bernard, R., Hofslundsengen, H., & Norbury, C., (2022). Anxiety and depression symptoms in children and adolescents who stutter: A systematic review and meta-analysis. Journal of Speech, Language & Hearing Research, 1.

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. I 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, these 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 difference 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 comorbidity 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., 2017b), 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, I 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 heterogeneous 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, I 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 Scale (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 years² (3) study assessed symptoms associated with DSM-classified ‘anxiety

² 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

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

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

Data Extraction

A coding scheme for extracting the relevant information about primary and additional outcomes was constructed and piloted (Higgins & Green, 2011) before data were independently extracted 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

(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, which are reported in the appendices for completeness.

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

Risk of bias of individual studies was assessed 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 $r = .50$).

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 3; see also *characteristics of excluded studies*, Appendix C). A total of 13 studies met the 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 Table 1 and Appendix D.

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).

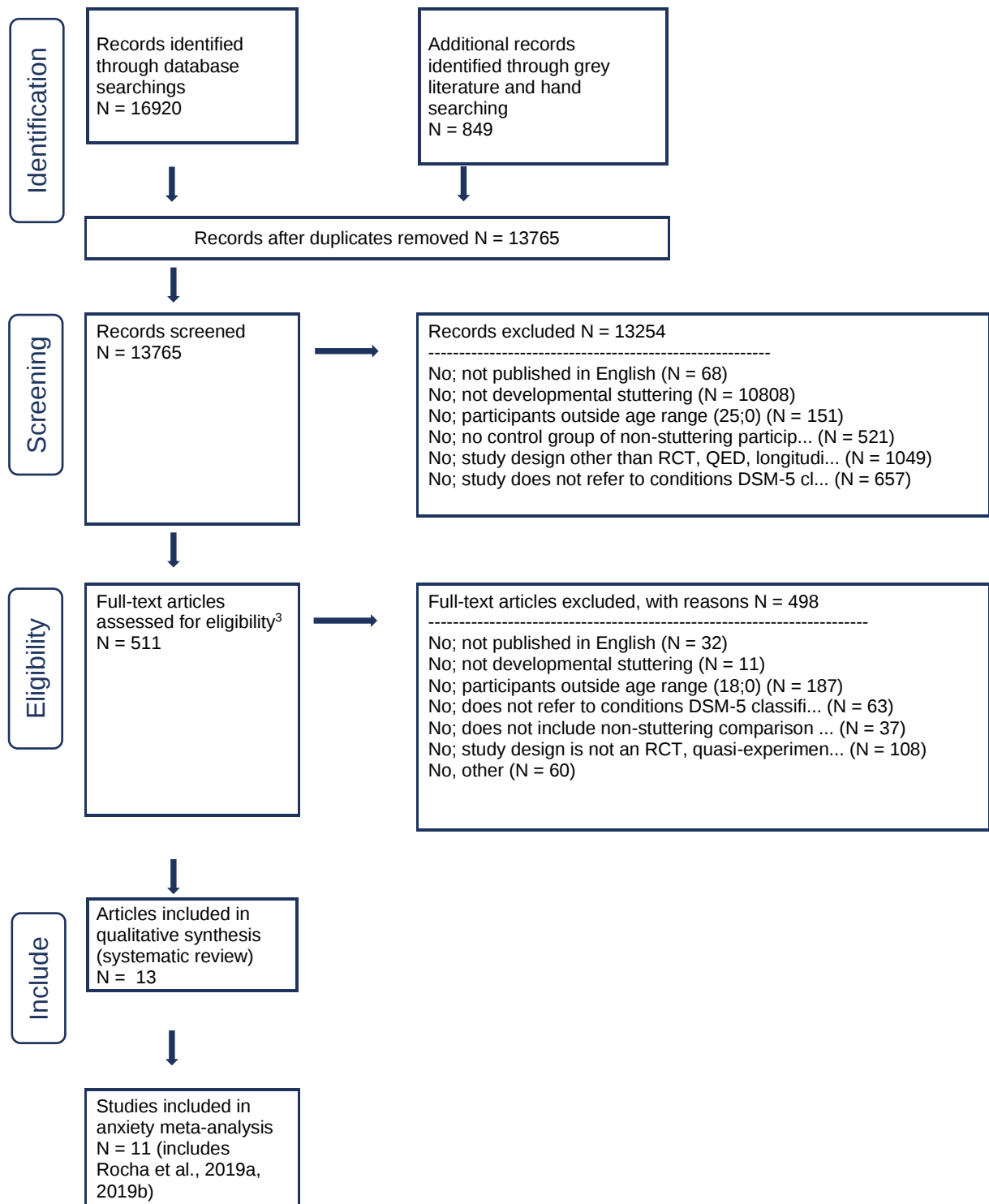


Figure 3: PRISMA flowchart outlining systematic review process (Distiller-SR)

³ The 10 articles identified in top-up search (2021) added here

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

Study Authors	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) 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) Reported correlation with state ($r = 0.115$) & trait anxiety ($r = 0.045$)	All received treatment previously but no treatment three months prior to the study	Recruited from those presenting for treatment
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	Inclusion criteria: no neurological or	English	Not reported	Iowa Scale: very mild (8); mild (8); mild-moderate		Suburban & metropolitan areas in five

	level 5 (bachelor degree). No significant difference between groups	intellectual impairment			(17); moderate (4); severe (5); very severe (4)		geographical areas, USA.
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%). 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. 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 No history of pharmacological intervention for anxiety-related disorders	Not reported Australian study	Not reported	%SS: mild (12%); moderate (47%); severe (41%) 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). 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). No significant correlation between RCMAS Total Score and SSI-4 ($r = -.09$)	Majority enrolled in speech therapy (none within last 6-months). Three never received treatment	SLTs & Psychologists identified potential families
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. 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% 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.
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SLT = speech and language therapist; min. = minimum; STAIC = State –Trait Anxiety Inventory for Children (Spielberger, 1973); S&L = speech and language; ADHD = attention-deficit/hyperactivity disorder.

* = studies met inclusion criteria for systematic review but excluded from meta-analysis

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 ($I^2 = 72.8\%$; $\text{Tau} = 0.40$) 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], $I^2 = 78.04\%$, $\text{Tau} = 0.41$).

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 78M : 18F 9–14yrs	104 59M : 45F 9–14yrs	State-Trait Anxiety Inventory for Children (STAIC)	Self-report	Australia	GENERAL <i>Trait anxiety measure</i>	-0.15 [-0.43 – 0.13]
Van der Merwe et al. (2011)	7 5M : 2F 3;3 – 4;11yrs	7 5M : 2F 3;2 – 4;10yrs	Pre-school Anxiety Scale (PAS)	Parent-report	New Zealand	<i>PAS total score</i>	-0.06 [-1.11 – 0.98]
						GENERAL <i>Generalised anxiety disorder subscale</i>	-0.27 [-1.32 – 0.79]
						SITUATION-SPECIFIC <i>Social phobia subscale</i>	0.00 [-1.05 – 1.05]
Rocha, Yaruss & Rato (2019a)	31 25M : 6F 7–9yrs	31 15M : 16F 7–9yrs	Multidimensional Anxiety Scale for Children (MASC) ¹ <i>Portuguese version</i>	Self-report	Portugal	GENERAL <i>MASC total score</i>	-0.06 [-0.55 – 0.44]
						SITUATION-SPECIFIC <i>Social anxiety subscale</i>	0.13 [-0.37 – 0.62]
Rocha, Yaruss & Rato (2019b)	19 11M : 8F 10–12yrs	19 7M : 12F 10–12yrs	Multidimensional Anxiety Scale for Children (MASC) ¹ <i>Portuguese version</i>	Self-report	Portugal	GENERAL <i>MASC total score</i>	0.15 [-0.49 – 0.79]
						SITUATION-SPECIFIC <i>Social anxiety subscale</i>	-0.20 [-0.84 – 0.43]

Hollister (2015)	46 36M : 10F 8–15yrs	46 36M : 10F 8–15yrs	The MacArthur Health & Behavioural Questionnaire 2.1 (HBQ 2.1)	Parent-report	USA	SITUATION-SPECIFIC <i>Social anxiety subscale</i>	0.26 [-0.15 – 0.68]
						GENERAL <i>General anxiety subscale</i>	0.54 [0.12 – 0.95]
Natarelli (2018)	19 14M : 5F 11–14yrs	19 Matched by sex, grade, ethnicity 11–14yrs	Revised Children's Manifest Anxiety Scale— Second Edition (RCMAS-2) <i>Italian version</i>	Self-report	Italy	GENERAL <i>RCMAS Total score</i>	0.38 [-0.27 – 1.02]
Iverach et al. (2016)	75 63M : 12F 7–12yrs	150 126M : 24F 7–12yrs	Spence Children's Anxiety Scale (SCAS)	Child-report (SCAS-C)	Australia	<i>SCAS total score</i>	SCAS-P: 0.51 [0.22 – 0.79]
				Parent-report (SCAS-P)			SCAS-C: 0.46 [0.18 – 0.74]
						GENERAL <i>Generalised anxiety subscale</i>	SCAS-P: 0.48 [0.20 – 0.76]
							SCAS-C: 0.32 [0.04 – 0.60]
						SITUATION-SPECIFIC <i>Social anxiety subscale</i>	SCAS-P: 0.57 [0.29 – 0.85]
							SCAS-C: 0.56 [0.28 – 0.84]
Davis et al. (2007)	18 16 M : 2F 10–16yrs	19 14M : 5F 10–15yrs	State-Trait Anxiety Inventory for Children (STAIC)	Self-report	England	GENERAL <i>Trait anxiety measure</i>	0.25 [-0.40 – 0.90]
						SITUATION-SPECIFIC <i>State anxiety measures</i>	Friends: 0.43 [-0.22 – 1.08]

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

Note. n = sample size, sex ratio and age range (years; months), male : female ratio; anxiety measure used in study; anxiety domain that measure was classified under ('situation-specific' = social anxiety and state anxiety scales; 'general' = generalised anxiety and trait anxiety scales); self- or parent-report measure; country in which study was undertaken; effect size (Hedge's g) with 95% confidence intervals. Rocha et al. (2019) divided their entire sample (n=100) into two age groups, which are reflected as younger (Rocha et al, 2019a) and older (Rocha et al, 2019b)

*Craig & Hancock (1996) compared state anxiety scores in the stuttering group with population norms and therefore state anxiety data were excluded from analysis.

¹RCMAS is considered a valid measure of chronic or trait anxiety (Reynolds, 1985); Multidimensional Anxiety Scale for Children (MASC) mainly taps trait anxiety (March et al., 1997), hence inclusion in the general anxiety domain.

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 anxiety⁴ and six social anxiety⁵ 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 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

4 Four of these state anxiety scores were from the same sample (Davis et al., 2007)

5 Includes both child- and parent-scores on the social anxiety subscale (Iverach et al., 2016)

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; Ntarelli, 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; Ntarelli, 2018) and the Multidimensional 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 categorised 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; Ntarelli, 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

Potential bias was investigated using funnel plots, Egger's Regression Test and the Failsafe N. Observation of the funnel plot (Figure 4) 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).

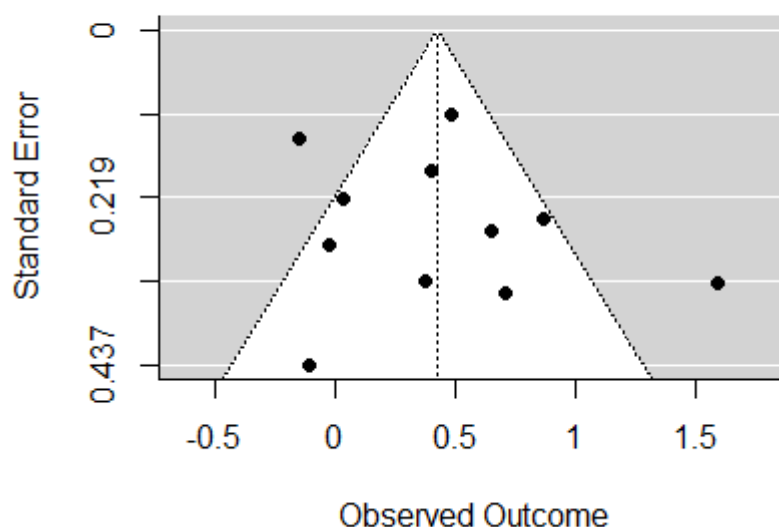


Figure 4: funnel plot displaying included studies

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

Notes. *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 it is not possible to 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 (Veerabhadrapa 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; Livingstone-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, Veerabhadrapa 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

My 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 I had aimed to assess anxiety and depression across a broad developmental period, only one pre-school study was included, 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 myriad ways. Given the limited number of studies available and the variety of symptom measures relied upon, the 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). I 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 I chose to group them separately. Consequently, it must be acknowledged that other researchers may choose to group these scales differently.

Clinical implications

These 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.

Chapter 3

Comparing anxiety and depression symptoms in children who do and do not stutter

Data in this chapter are reported in a registered pre-print (not peer-reviewed): Bernard, R., Griffiths, S. L., & Norbury, C. (2022, August 16). Comparing anxiety and depression symptoms in children who do and do not stammer. <https://doi.org/10.31234/osf.io/jkud9>

Abstract

Purpose. Children who stutter may be at increased risk of anxiety compared to non-stuttering peers. Although anxiety and depression commonly co-occur, there are few studies evaluating the association between childhood stuttering and depression symptoms. The purpose of this study was to investigate whether children who do and do not stutter differ significantly in anxiety and depression symptoms, and the extent to which this association may vary depending on age and on the respondent.

Methods. Forty-six children who stutter (8 – 15 years) and 46 children who do not stutter (10 – 14 years) and their parents, completed the Revised Children's Anxiety & Depression Scale (RCADS-25). To determine whether groups differed significantly on anxiety and depression subscale scores, multiple regression models were fitted to the data, accounting for group differences in age and socio-economic status (SES). Intra-class correlation coefficients were calculated to assess parent-child agreement on each subscale.

Results. Self-reported anxiety and depression symptoms increased with age in the stuttering group but not in the comparison group. After accounting for age and SES, parents reported significantly higher anxiety, but not depression, symptoms for children who stutter. However, very few participants scored above clinical threshold on either subscale. Good agreement was found between parent- and child-reported symptom scores for the stuttering group. Comparison of two community samples found no

differences in likelihood of scoring above clinical threshold for anxiety or depression.

Conclusion. Children who stutter may experience increasing anxiety and depression symptoms with age, although these findings need replication in larger, longitudinal studies. These findings underline the need for further research into internalising symptoms in children who stutter across development, and consideration of factors that may be associated with elevated risk for this population.

Introduction

Stuttering is a neurodevelopmental condition characterised by disruptions in spoken fluency. In addition to core speech behaviours, stuttering may also be associated with accessory features, for example muscle tension, and emotional states, such as negative feelings and attitudes (Guitar, 2014; Wingate, 1964). Stuttering affects approximately 5% to 8% of children during development with onset typically occurring between two and five years of age, although later onset may occur up to adolescence (Yairi & Ambrose, 2013; Guitar, 2014; Howell, 2011).

Spoken communication is central to social interaction and societal participation. Disruptions to spoken fluency in conversation, as well as physical behaviours observed in some people who stutter may contribute to negative feedback from communication partners and potentially lead to a child withdrawing from social situations and becoming increasingly isolated. Research has shown that as early as pre-school, stuttering has been associated with negative peer reactions and negative effects on interaction with other children (Langevin, Packman & Onslow, 2009).

Low self-perceived communication competence, heightened communication apprehension and fear of negative evaluation have been reported in adolescents who stutter (Erickson & Block, 2013; Mulcahy et al., 2008; Blood, Blood, Tellis & Gabel, 2001). Furthermore, children who stutter are at increased risk of bullying and peer rejection (Blood et al., 2011; Blood & Blood, 2007; Davis, Howell & Cooke, 2002). Childhood bullying may be associated with anxiety, depression and low self-esteem in the stuttering population (Cook & Howell, 2014; Blood & Blood, 2007; Hugh-Jones & Smith, 1999). Consequently, the experiences of children and adolescents who stutter may put them at greater risk of adverse mental health outcomes relative to non-stuttering peers (Smith et al., 2014).

Anxiety in children and adolescents who stutter

The most recent Mental Health in Children and Young People Survey (2017) published prior to the COVID-19 pandemic, indicated that emotional disorders (including anxiety, depression and bipolar affective disorders) were the most prevalent category of mental health disorder among children and adolescents (5 – 19 years) in England (Sadler et al., 2018). Anxiety disorders were more common (7.2%) than depressive disorders (2.1%) across 5 – 19 year olds, but rates increased with age, especially in girls. Prior to the COVID-19 pandemic, the prevalence of mental health disorders among children and adolescents in the UK had been increasing (Pitchforth et al., 2018).

Anxiety is an emotional state, which generates cognitive, behavioural and physiological responses to perceived threat (Essau, 2007). The number, persistence, severity and functional impairment of symptoms differentiate normal levels of anxiety experienced by all children from pathological anxiety (APA, 2013; Essau, 2007). The DSM-5 (APA, 2013) cites anxiety around speaking in its diagnostic criteria for childhood onset fluency disorder (stuttering). The adult literature indicates that stuttering is associated with increased risk of anxiety symptoms and social anxiety disorder (Craig & Tran, 2014; Iverach & Rapee, 2014). However, research evaluating anxiety and depression in children who stutter has produced variable findings. A recent systematic review and meta-analysis found elevated symptoms of anxiety in some children who stutter, with a moderate effect size difference in anxiety symptom scores between children who do and do not stutter (Bernard, Hofslundsengen & Norbury, 2022). However, substantial between-study variance was also reported, suggesting heterogeneity in symptom reporting. Earlier narrative reviews concluded there was limited evidence of elevated anxiety amongst young children who stutter but hypothesised that anxiety symptoms may increase with age (Smith et al., 2014; Alm, 2014). Age of onset for anxiety during childhood and adolescence remains unclear, with many studies comprising samples spanning wide age ranges (Smith et al., 2014).

Several methodological issues have been cited as possibly contributing to inconsistent findings, including small sample sizes and insufficient statistical power, selection bias, failure to control for potential moderating factors such as co-occurring difficulties and treatment status, utility of a multitude of assessment measures tapping differing anxiety constructs, and limited sensitivity of psychological measures for this clinical population (Bernard et al., 2022; Iverach et al., 2011; Jones et al., 2021; Menzies et al., 1999; Smith et al., 2014). The validity of conclusions might be undermined by reliance on clinically ascertained samples, given that individuals seeking treatment may be more likely to present with elevated anxiety, while recent therapy for stuttering may positively influence mental health outcomes (Craig et al., 2003; Craig, 1994). Community studies have found little evidence of elevated anxiety in school-aged children who stutter compared to non-stuttering peers (Andrews & Harris, 1964; Smith et al., 2017).

While many studies have focused on anxiety symptoms, often measured through self- or parent-report, others have captured prevalence of clinical disorder in children and adolescents who stutter through in-depth diagnostic assessment. For instance, Iverach et al. (2016) used an online diagnostic assessment for diagnosis of DSM-IV (APA, 2000) anxiety disorders and found significantly higher prevalence of social anxiety disorder among children (aged 7 – 12years) who stutter compared to non-stuttering children, with six-fold increased odds of meeting diagnostic criteria for anxiety. Gunn et al. (2014) reported that 38% of adolescents who stutter met criteria for an Axis-I mental health disorder (DSM-IV; APA, 2000), the majority of which involved anxiety disorders at a rate that was substantially higher than the prevalence in the general population. However, both of these studies involved participants who were seeking or receiving treatment for stuttering. While these studies contribute to our understanding of the needs and profiles of children who access services, it is also important to consider the population prevalence of anxiety and depression in children who stutter, including those who do and do not access intervention.

The association between depression symptoms and stuttering

Community studies have found that depression is often preceded by anxiety in adolescents (Lewinsohn et al., 1997; Stein et al., 2001), which further supports the need to consider both conditions in the stuttering population. Despite common co-morbidity between anxiety and depression amongst children and adolescents in the general population (Lewinsohn et al., 1997; Seligman & Ollendick, 1998; Stein et al., 2001), there appears to be limited research investigating depression in the stuttering population.

In a large cohort study of adolescents seeking treatment for stuttering, Iverach et al. (2017) found mean anxiety and depression scores fell within normal limits, although higher anxiety scores predicted higher depression scores. Recent reviews found little evidence of elevated symptoms of depression in children and adolescents (<18 years) who stutter compared to fluent peers (Bernard, Hofslundsengen & Norbury, 2022; Livingstone-Pountney & Mitrevski, 2019). However, some studies have reported elevated depression symptoms in young adults who stutter (Briley, Gerlach & Jacobs, 2021; Doruk et al., 2008; Livingstone-Pountney & Mitrevski, 2019), perhaps indicating risk for depression occurs later in development. Given the high comorbidity between anxiety and depression, and evidence suggesting that at least some children and adolescents who stutter report increased symptoms of anxiety relative to non-stuttering peers, further investigation of depression symptoms is warranted.

Respondent differences in symptom reporting

Poor correspondence between informant ratings of social, emotional and behavioural problems, internalising symptoms, and quality of life have been widely documented in the literature (De Los Reyes & Kazdin, 2005; Achenbach, McConaughty & Howell, 1987; Duhig et al., 2000; Upton, Lawford & Eiser, 2008). In their critical review, De Los Reyes and Kazdin (2005) cite evidence of low to moderate agreement between informants on ratings of child anxiety and child depression.

Previous research has shown that parents tend to perceive stuttering as negatively affecting their children's quality of life, notably in terms of self-esteem, mood and well-being (Guttormsen, Yaruss & Næss, 2021). When examining the impact of stuttering as perceived by children and parents, low correlations between ratings have been reported in young and school-aged children (Ntourou et al., 2017; Vanryckeghem, 1995), although a recent study of Portuguese children who stutter found broadly similar parent and child ratings of overall impact of stuttering (Rocha, Yaruss & Rato, 2020). Studies examining factors that may be associated with parent perceptions of stuttering impact suggest an association with positive family history of stuttering, higher stuttering severity, and shorter time since onset (Ntourou, et al., 2017; Rocha et al., 2020; Wheeler et al., 2011). However, Guttormsen et al. (2021) found no differences in parent impact ratings based on children's therapy status, age, sex or family history of stuttering, nor any association between impact ratings and parent ratings of stuttering severity or frequency. Nevertheless, the negative parental perception of stuttering on children's wellbeing may contribute to differences in symptom reporting between children and parents.

Studies examining anxiety and stuttering in the literature to date have utilised both parent- and self-report measures. It may be that respondent effects have contributed to some studies reporting evidence of elevated symptoms, and others not. As discussed in chapter two, of the 11 studies included in the systematic review, eight involved self-report, two parent-report and one involved both. Iverach et al. (2016) found parents reported significantly lower anxiety scores on the Spence Children's Anxiety Scale (SCAS; Spence, 1998) relative to school-aged children (7 – 12 years) who stutter seeking speech treatment. In order to examine respondent effects on symptom reporting, measures that involve self- and parent-report are required. Few studies have directly compared self- and parent-reported scores on the same measure, necessitating the need for further examination of respondent effects in this population. In the present study, it was hypothesised that parents would report higher symptom scores than children because parents may worry about the impact of speech dysfluency on their child.

The present study

The present study extends previous investigations concerning risk for elevated anxiety and depression symptoms in children who stutter compared to those who do not in a community sample. The study was initiated prior to the Covid-19 pandemic and data collection has been affected by school closures. Data collected from children who stutter and children who do not stutter during the period of coronavirus restrictions in the UK will be analysed to consider the following research questions:

- Do children who stutter have elevated symptoms of anxiety and/or depression compared with children who do not stutter?
- Do a higher proportion of children who stutter score above threshold on an overall measure of anxiety and depression, relative to children who do not stutter?
- Are high anxiety scores associated with higher depression scores in children who stutter?
- Is there a significant difference in parent-reported and child-reported anxiety and depression scores in children who stutter?

Method

This study was pre-registered on the Open Science Framework ([10.17605/OSF.IO/4FGEX](https://osf.io/4FGEX)) in October 2019, prior to the onset of Covid-19. Subsequent amendments were made to the planned analyses in March 2021, prior to data analysis taking place, to account for changes in recruitment and data collection strategies that resulted from school closures. Ethical approval was granted by University College London (UCL) Research Ethics Committee (15535/001). Parental consent and child assent were obtained from participants at the beginning of the questionnaire.

Participants

Data for the stuttering group were collected using an online questionnaire available between February 2020 and August 2021. The comparison group

comprised a sub-sample of participants taking part in the longitudinal Surrey Communication and Language in Education Study (SCALES; Norbury et al., 2016), who had participated in an online or in-person testing session over approximately the same period.

The primary objective of the present study was to establish whether or not children who stutter report elevated symptoms of anxiety and/or depression relative to non-stuttering peers. In an effort to address the potential confounding influence of the pandemic on mental health symptoms reported during the COVID-19 global pandemic, two changes were made to the pre-registered protocol. Firstly, only SCALES data collected over approximately the same period as the online questionnaire were included in analyses, rather than comparing scores with population norms. Consequently, data used in all analyses were collected in slightly different ways. Secondly, a time variable was created indicating whether data had been collected prior to the pandemic, during the pandemic or during a period of national lockdown. This was used to aid matching of participants, alongside other demographic data. It should be noted that all data are cross-sectional, providing a snapshot of symptoms for each group over this period. Hence, this study design does not permit speculation as to the changing patterns of symptom presentation or severity relating to COVID-19.

Stuttering Group

(i) Recruitment

Children who stutter and their parents were invited to take part in an online questionnaire, which was promoted through a UK stuttering charity and social media. To maximise recruitment from a diverse population, the study was advertised to organisations that speech and language therapists and families of children who stutter are likely to engage with in the UK. A flyer about the study was sent to schools across London and the South East of England.

(ii) *Inclusion criteria*

Inclusion criteria to take part in the questionnaire were as follows: (i) child has a developmental stutter; (ii) child is aged between 8 and 13 years; (iii) child lives in the UK. Five participants who completed the questionnaire fell outside this age range. As the Revised Children's Anxiety and Depression Scale (RCADS-25) is normed on children aged 8 – 18 years, data for children under eight were removed ($n = 3$). The two older participants (aged 14 and 15) were retained in the analyses. The questionnaire was open to children living in all four nations of the UK.

(iii) *Sample characteristics*

The stuttering group comprised 46 children and adolescents who stutter. Developmental stuttering was determined by parent-report. Demographic information (age, sex and socio-economic status) was unavailable for one participant. The stuttering group comprised 12 female and 33 male participants (one undisclosed), and the mean age was 130 months ($SD = 22$). All participants spoke English as their first language, and eight participants reported speaking an additional language. The majority of participants who completed the questionnaire lived in England, while four respondents resided in the devolved nations of the UK.

The Income Deprivation Affecting Children Index (IDACI) was used as an indicator of family socio-economic status (SES). The IDACI is one of the supplementary indices of the English Indices of Deprivation (IoD), which measures the proportion of children (0 – 15 years) living in deprived families and is based on home postcode⁶. The IoD ranks all neighbourhoods in England from 1 (most deprived) to 32,844 (least deprived). The mean IDACI rank for the stuttering group was 16,849 ($SD = 8,594$), which is indicative of the 50% – 60% most deprived neighbourhoods. Each country in the UK publishes its own indices of multiple deprivation, which are not directly comparable because of differences in the indicators used, time periods

⁶ Ministry of Housing, Communities & Local Government, English Indices of Deprivation (2019). [English indices of deprivation 2019 - GOV.UK \(www.gov.uk\)](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/812222/english-indices-of-deprivation-2019.pdf) [accessed 16.11.21]

measured and the size of areas⁷. For those living outside of England, it was not possible to calculate the IDACI rank as a UK-wide version is unavailable. SES data is therefore unavailable for seven participants due to non-disclosure or reportedly living in one of the devolved nations.

Family background information was available for all 46 participants (Table 4). The reported age of stuttering onset ranged between two and seven years ($M = 3.52$, $SD = 1.41$). All participants reported that they had accessed Speech and Language Therapy. Most participants ($n=40$) confirmed that they had been diagnosed with a stutter by a speech and language therapist, and half of participants reported a family history of stuttering ($n=23$). Almost half of participants ($n=22$) reported a family history of mental health difficulties, while six respondents reported that their child had accessed support for mental health previously.

To evaluate self-perceived stuttering severity, children were asked to rate the severity of their stutter on a non-standardised rating scale (1 = not very severe/bumpy; 10 = very severe/bumpy). The mean self-perceived stuttering severity rating was 6 ($SD = 1.95$, $range = 3 - 10$, $n=35$).

Comparison Group

The non-stuttering comparison group comprised 46 children and adolescents who were part of the longitudinal Surrey Communication and Language in Education Study (SCALES). The SCALES study is a UK population study of language development that followed 590 children from age 5 to 13 years. Children were recruited to this study from schools in Surrey, England. Further information about the study design employed in the SCALES population study can be found in Norbury et al. (2016). All children in the SCALES study who had complete child and parent responses on the RCADS-25 collected between 2019 and 2021, were identified as possible

⁷ Ministry of Housing, Communities & Local Government. The English Indices of Multiple Deprivation 2019. Research Report.
https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/833947/loD2019_Research_Report.pdf [accessed 16.11.21]

matches to the participants in the stuttering group. Participants were then matched as closely as possible on lockdown status, IDACI rank, sex and age with participants in the stuttering group. Further information about this process is included below. For the purposes of comparison with the stuttering group in the present study, child and parent responses to the RCADS-25, demographic and background information were extracted for analyses.

Demographic data (sex, socio-economic status, age) were available for all 46 participants in the comparison group. The mean age of participants was 156 months ($SD = 14.2$) and included 17 female and 29 male participants. The mean IDACI rank was higher than for the stuttering group ($M = 23,309$, $SD = 7,031$), indicating the comparison children lived in less deprived areas (70% - 80% least deprived neighbourhoods).

Complete family background information was available for 39/46 participants (Table 4). Of the data available, all but one child spoke English as their first language ($n=38$). Ten participants reported a positive family history of mental health difficulties, and six children had accessed Child and Adolescent Mental Health Services (CAMHS). Seven participants had accessed speech and language therapy previously and two were accessing intervention at the time of data collection. A total of 11 participants had a clinical diagnosis, such as a neurodevelopmental condition, sensory impairment or genetic disorder. Two participants reportedly had more than one co-occurring condition. Parents of two further participants reported difficulties that were not yet formally diagnosed.

Measures

(i) *Revised Children's Anxiety and Depression Scale (RCADS-25)*

The RCADS-25 (Ebesutani et al., 2012) is a self-report measure that assesses the frequency of anxiety and low mood symptoms in children and adolescents between 8 and 18 years of age. It is a shortened version of the 47-item Revised Children's Anxiety and Depression Scale (RCADS; Chorpita

et al., 2000), which is one of the Child Outcome Research Consortium (CORC) measures. In an effort to reduce the burden on participants and optimise likelihood of questionnaire completion, the RCADS-25 was selected over the full RCADS. Use of the RCADS-25, as opposed to the RCADS, has been advocated when the objective is to obtain an estimate of general anxiety and depression problems using a briefer yet reliable tool (Ebesutani et al., 2012). The RCADS-25 is included in the standard set of clinical outcome measures for child and adolescent anxiety and depression symptoms agreed by an International Consortium (ICHOM; Krause et al., 2021). The Wellcome Trust⁸ and National Institute of Mental Health⁹ recommend the RCADS-25 for research into anxiety and depression in children and adolescents.

There are parent and child versions of the RCADS-25 (RCADS-25-C, Ebesutani et al., 2012; RCADS-25-P, Ebesutani et al., 2017), both of which provide three scores: 'Total Anxiety', 'Total Depression' and 'Total Anxiety & Depression'. They retain the 10-item Depression Scale from the full version, while the Anxiety Scale comprises 15 items based on the five subtypes of anxiety measured in the full RCADS, providing a measure of broad anxiety (Ebesutani et al., 2012). Participants receive a score (0-3) corresponding to the response (never; sometimes, often, always) selected for each of the 25 statements. Higher scores indicate greater symptom severity. Raw scores can be converted to T-scores, which indicate whether symptom scores fall above or below the clinical cut-off for this measure. The RCADS-25-C also illustrates good discriminatory validity for anxiety and depression. Both the Depression Scale and Anxiety Scale have good reliability in clinical ($\alpha = 0.80$, $\alpha = 0.91$ respectively) and school ($\alpha = 0.79$, $\alpha = 0.86$ respectively) samples (Ebesutani et al., 2012). The RCADS-25-P anxiety and depression subscales similarly show good reliability ($\alpha = .80 - .90$) in both school and clinical populations (Ebesutani et al., 2017).

8 Wellcome Trust, Common Metrics in Mental Health Research [accessed 3.4.23]:

<https://wellcome.org/grant-funding/guidance/common-metrics-mental-health-research>

9 National Institute of Mental Health, Notice Announcing the National Institute of Mental Health (NIMH) Expectations for Collection of Common Data Elements [accessed 3.4.23]: <https://grants.nih.gov/grants/guide/notice-files/NOT-MH-20-067.html>

Although normed on a US population, the RCADS-25 facilitates evaluation of both anxiety and depression symptoms within one measure and provides insight into multi-informant symptom reporting. Consequently, through selection of the RCADS-25 this research contributes to two research gaps in the field: respondent effects on symptom reporting and co-morbidity of anxiety and depression in stuttering. A key consideration when involving children in research is balancing the quantity and quality of data collection with time burden. The RCADS-25 can be completed in a short amount of time, an important advantage given the correlation between survey length and participation in health research (Booker, Austin & Balasubramanian, 2021).

(ii) *Online questionnaire*

The online questionnaire was created using Qualtrics software (Qualtrics, Provo, UT) and was only completed by the stuttering group. It was divided into two parts: one section for the parents/guardians and one for the child. In the parent section, participants were asked to provide background information about the child and then complete the RCADS-25 parent version (RCADS-25-P). The child section involved completion of the RCADS-25 child version (RCADS-25-C), the Illinois Bullying Scale (IBS; Espelage & Holt, 2001) and a stutter-severity rating on a 9-point Likert scale. Data obtained from the IBS are reported in chapter four. Children were offered a National Book Token to thank them for their participation.

(iii) *SCALES data*

Children in the comparison group completed a battery of language activities either in person or online with a trained researcher. The RCADS-25-C was completed online using Qualtrics software. Families were offered the option of an online or paper version of the RCADS-25-P. Only demographic data, family background information and responses to the RCADS-25 child and parent versions were included in the present analyses.

Analyses

All analyses were performed in R version 4.0.3 (R Core Team, 2020).

Participants were matched using the *ccoptimalmatch* package (Mamouris & Nassiri, 2021) in R based on sex, age, IDACI rank and lockdown status.

Participants were first matched on lockdown status (100% match) and then matched as closely as possible on age, sex and IDACI rank. The following analyses were conducted to address the four hypotheses:

- (1) There will be a significant difference in mean scores (RCADS-25) between children who stutter and the comparison group.

The primary outcome was the mean difference in RCADS-25 anxiety and depression subscale scores between the stuttering and non-stuttering comparison group. Four dependent variables were included in the analyses: child-reported anxiety and depression subscale scores, and parent-reported anxiety and depression subscale scores. RCADS-25 Total Scores are calculated by summing together the anxiety and depression subscale scores and are reported but not included in subsequent analyses. To investigate whether symptom scores differed between groups, regression models for the four dependent variables were fitted. Although in the pre-registration I had planned to analyse data using *t*-tests, the groups differed significantly on two factors (age and SES) associated with adverse mental health (Fryers, Melzer & Jenkins, 2003; Pine & Klein, 2015; Thapar et al., 2012). Therefore, regression models were fitted to evaluate mean differences in subscale scores between groups, while accounting for these differences. First, I ran unadjusted regression analyses with group as a predictor and RCADS-25 subscale scores as the outcome (these are equivalent to running *t*-tests). I then ran adjusted analyses which included age and SES variables as predictors to evaluate the effect of group once these variables were held constant. I have reported the results of both unadjusted and adjusted models for each of the four subscales.

- (2) Relative to the comparison group, a higher proportion of children who stutter will score above clinical threshold on an overall measure of anxiety and depression.

Raw scores on the RCADS-25 can be converted into T-scores, which account for US grade and sex of the child and indicate whether scores fall above or below clinical threshold. T-scores can be interpreted as indicative of low severity (<65), borderline clinical threshold (65 – 70) and above (>70) clinical threshold (Ebesutani et al., 2012). The RCADS-25 is normed on a US population and consequently T-scores should be interpreted with caution. In the stuttering group, I estimated participants' US school grade based on the child's age at the approximate date of questionnaire completion. T-scores were calculated for participants in the stuttering ($n=34$) and comparison ($n=46$) groups. Due to missing demographic data ($n=1$) or missing child-reported scores ($n=11$), T-scores were unavailable for a total of 12 children who stutter. Fisher's Exact Test assessed the association between group and frequency of scoring above or below clinical threshold on anxiety and depression subscales. I have calculated T-scores to address this second hypothesis, however raw scores have been used in all other reported analyses due to the limitations of using pre-pandemic norms based on a non-UK population.

- (3) Children who stutter who score highly on the anxiety subscale will also score highly on the depression subscale.

Pearson's Correlation Coefficients estimated the association between anxiety and depression scores in each group.

- (4) There will be a significant difference in parent-reported and child-reported mean scores on the RCADS-25 for the stuttering and comparison groups.

Finally, to evaluate whether there were significant differences in mean subscale scores by respondent (self- or parent-report), I initially performed four two sample *t*-tests. To account for multiple comparisons, the Bonferroni correction was applied using the *p.adjust* function in R. In exploratory analyses, inter-rater reliability was assessed in each group by calculating the intraclass correlation coefficient (ICC) using the *irr* package in R. ICC values may indicate poor (<0.5), moderate (0.5 – 0.75), good (0.75 – 0.9) or excellent (>0.9) reliability (Koo & Li, 2016). While Pearson's correlation

coefficient provides information about the strength of association between values, it cannot capture agreement between raters (Bland & Altman, 1986; Stolarova et al., 2014).

Results

In the stuttering group, child-reported scores were available for 35 participants; parent scores were available for all participants ($n = 46$). Participants in the comparison group contributed both child and parent scores on the RCADS-25 ($n = 46$). Table 4 summarises the sample characteristics for each group. The mean age difference between matched pairs was just over one year ($M = 13.28$ months, $SD = 19.50$). The mean difference in IDACI rank between pairs was 5,011 ($SD = 7,380$). In both the stuttering and comparison groups, there were more boys ($n = 33$, $n = 29$ respectively) than girls ($n = 12$, $n = 17$ respectively). Pearson's Chi Square Test indicated there was no significant association between sex and group ($\chi^2(1, N = 91) = 1.11$, $p = 0.29$), suggesting that participants were reasonably matched on sex. However, two sample t -tests indicated there were significant differences between groups in both the mean age of participants ($t(72.92) = -6.64$, 95% CI $[-33.87, -18.22]$, $p < .001$) and the mean SES ($t(73.40) = -3.75$, 95% CI $[-9894.19, -3027.04]$, $p < .001$), reflecting the comparatively lower mean age and lower SES of the stuttering group.

Anxiety and depression scores

Table 5 shows the mean total and subscale scores on the RCADS-25 for both groups by respondent. Table 6 presents child-reported scores for each subscale by sex and group. In the stuttering group, mean scores suggest girls report fewer depression symptoms than boys, but mean scores were similar for anxiety. In contrast, mean scores for the comparison group suggest girls report more symptoms of both anxiety and depression, on average, than boys. All analyses have been conducted using the entire sample for each group due to small sample sizes when separating groups by sex.

Table 4: Child demographic and family characteristics for the stuttering and comparison groups. *M*(*SD*) for continuous variables, *N* (%) for categorical variables

		Stuttering group (N = 46)	Comparison group (N = 46)	Group differences
Demographic information ^a				
Age in months <i>M</i> (<i>SD</i>)		130 (22) 96 – 191	156 (14) 125 – 178	$t(72.92) = -6.64^*$
Sex <i>N</i> (%)	Female	12 (26%)	17 (37%)	$\chi^2(1, 91) = 1.11$
	Male	33 (72%)	29 (63%)	
IDACI rank <i>M</i> (<i>SD</i>)		16,849 (8,594)	23,309 (7,031)	$t(73.40) = -3.75^*$
Child & family characteristics ^b				
First language (English) <i>N</i> (%)	Yes	46 (100%)	38 (82.6%)	
	No	-	8 (17.4%)	
Clinical diagnoses <i>N</i> (%)	Yes	-	11 (24%)	
	No	-	35 (76%)	
Access to speech & language therapy^c <i>N</i> (%)	Yes	46 (100%)	9 (19.6%) ^d	
	No	-	37 (80.4%)	
Family history of mental health <i>N</i> (%)	Yes	22 (47.8%)	10 (21.7%)	
	No	24 (52.2%)	36 (78.3%)	
Child previously accessed support for mental health ^e <i>N</i> (%)	Yes	6 (13%)	6 (13%)	
	No	40 (87%)	40 (87%)	
Diagnosis of stuttering confirmed <i>N</i> (%)	Yes	40 (87%)	-	
	No	6 (13%)	-	
Approximate age of onset (years) of stuttering <i>M</i> (<i>SD</i>)		3.52 (1.41)	-	
Family history of stuttering <i>N</i> (%)	Yes	23 (50%)	-	
	No	23 (50%)	-	

Note.

^a Complete demographic information available for 45/46 participants in stuttering group. Additional six participants in stuttering group did not have SES data. Complete demographic information available for 46/46 participants in comparison group.

^b For child & family characteristics, information available for 46/46 participants in stuttering group. Child & family characteristic data available for 39/46 in the comparison group.

^c Families in the stuttering group were asked whether their child had accessed Speech & Language Therapy for assessment/therapy for stuttering, whereas the comparison group were asked whether their child had previously or were currently accessing Speech & Language Therapy.

^d Previous intervention (n=7), current intervention (n=2)

^e The stuttering group were asked whether the child had ever accessed support from Child & Adolescent Mental Health Services (CAMHS) or psychological services; the comparison group were asked whether the child had accessed CAMHS.

*p<.001

Table 5: Mean raw scores (SD) and range for total and subscale scores on the RCADS-25-C and RCADS-25-P for each group

RCADS-25 subscale	Respondent	Stuttering group M (SD) Range	Comparison group M (SD) Range
Total scores	Child-report	15.54 (8.83) 1 – 34	17.78 (10.57) 4 – 52
	Parent-report	12.63 (8.01) 1 – 37	9.57 (7.29) 0 – 35
Anxiety subscale	Child-report	9.74 (5.93) 1 – 23	9.93 (6.43) 1 – 28
	Parent-report	7.57 (5.11) 1 – 23	5.46 (4.15) 0 – 21
Depression subscale	Child-report	5.80 (3.92) 0 – 16	7.85 (4.63) 1 – 24
	Parent-report	5.07 (3.54) 0 – 15	4.11 (3.93) 0 – 19

Note. Stuttering group: child-report, n=35; parent-report, n = 46. Comparison group: n=46 for child and parent report. Scoring range for RCADS-25: Total score (0 – 75), Anxiety subscale (0 – 45), Depression subscale (0 – 30)

Table 6: Mean scores by sex for child-reported total and subscale scores in the stuttering ($n = 35$) and comparison ($n = 46$) groups

RCADS-25 scale	Stuttering group		Comparison group	
	Female ($n=12$)	Male ($n=33$)	Female ($n=17$)	Male ($n=29$)
Total score	14.33 (10.55)	16.08 (8.50)	19.94 (9.34)	16.52 (11.19)
Anxiety subscale score	9.89 (7.82)	9.68 (5.43)	11.47 (6.15)	9.03 (6.53)
Depression subscale score	4.44 (3.13)	6.40 (4.14)	8.47 (3.73)	7.48 (5.11)

Data for each variable were visualised using histograms and boxplots; data for some variables were skewed and not normally distributed. Inspection of the boxplots for each variable indicated presence of 11 potential outlier scores associated with seven participants (two in the stuttering group and five in the comparison group). Most outliers were identified among parent-reported scores ($n = 8$) relative to child-reported scores ($n = 3$). In the stuttering group, parent-reported scores on both subscales were identified as outliers for two participants. All but one of these 11 extreme scores were associated with T-scores that fell above clinical threshold. Given that data were collected from two community samples, I had anticipated scores across the entire range of the RCADS-25 and for scores to be skewed to the left (i.e. higher proportion of lower scores) than perhaps would have been expected in a clinical sample receiving treatment for mental health difficulties. As the purpose of this study was to ascertain whether children who stutter show evidence of elevated anxiety symptoms relative to children who do not stutter, potential outliers were considered meaningful and therefore retained in all analyses. Violin plots, which illustrate the distribution and density of scores for each group, can be found in Appendix E.

Regression Analyses

Multiple regression models were fitted to the data to investigate whether children who stutter report higher subscale scores than children who do not stutter. Results of unadjusted and adjusted regression analyses are provided for child-reported (Table 7) and parent-reported (Table 8) subscale scores.

Child-reported scores

Anxiety subscale scores

Unadjusted results indicated that there was no significant effect of group on child-reported anxiety scores ($b = 0.19$, $SE = 1.40$, $t = 0.14$, $p = .89$). This model also explained very little of the variance in anxiety scores ($F(1, 79) = 0.02$, $p = .89$, adj. $R^2 = -0.012$). When age and SES were entered into the model, there was an interaction between age and group ($b = -0.30$, $SE = 0.08$, $t = -3.69$, $p = <.001$). This second model was shown to be a significant fit to the data, accounting for 12.9% of the variance in child-reported anxiety scores ($F(4, 71) = 3.78$, $p = .008$, adj. $R^2 = 0.129$).

Given the interaction between age and group, follow-up analyses were performed to test whether there was a main effect of age on anxiety scores in each group separately. Regression models were run with age and SES entered as predictors. In the stuttering group, there was a significant main effect of age on anxiety scores in so far as for every one month increase in age, there was a 0.15 point increase in self-reported anxiety scores ($b = 0.15$, $SE = 0.05$, $t = 2.91$, $p = .007$). In contrast, for the comparison group the significant main effect of age on anxiety scores was characterised by a 0.15 point decrease in score for every one month increase in age ($b = -0.15$, $SE = 0.06$, $t = -2.398$, $p = .02$). There was no effect of SES on anxiety score in either group. Figure 5 illustrates that for the stuttering group, as age increases so do anxiety scores, however for the comparison group, anxiety scores decrease with increasing age.

Depression subscale scores

Results of the unadjusted analyses suggested there was a marginally significant effect of group for child-reported depression scores ($b = 2.05$, $SE = 0.97$, $t = 2.10$, $p = .04$), as the comparison group reported higher depression symptoms before accounting for age and SES differences. This model explained only a small amount of the variance ($F(1, 79) = 4.43$, $p = .04$, adj. $R^2 = 0.041$). In the adjusted analyses, there was a significant interaction between age and group ($b = -0.22$, $SE = 0.05$, $t = -4.05$, $p <.001$).

This adjusted model was a significant fit to the data ($F(4, 71) = 5.57, p = .001$, adj. $R^2 = 0.1959$), accounting for approximately 19.6% of the variance in child-reported depression scores.

Follow-up analyses indicated a significant main effect of age on self-reported depression scores in both the stuttering and comparison groups. For the stuttering group, results of regression analyses indicated that for every one month increase in age, there was a 0.09 point increase in depression score ($b = 0.09, SE = 0.03, t = 2.66, p = .01$). However, in the comparison group, a one month increase in age was associated with a 0.14 point decrease in self-reported scores ($b = -0.14, SE = 0.04, t = -3.07, p = .004$). There was no effect of SES on depression score in either group. Figure 6 shows that in the stuttering group, child-reported scores increased with age, whereas in the comparison group scores decreased with age.

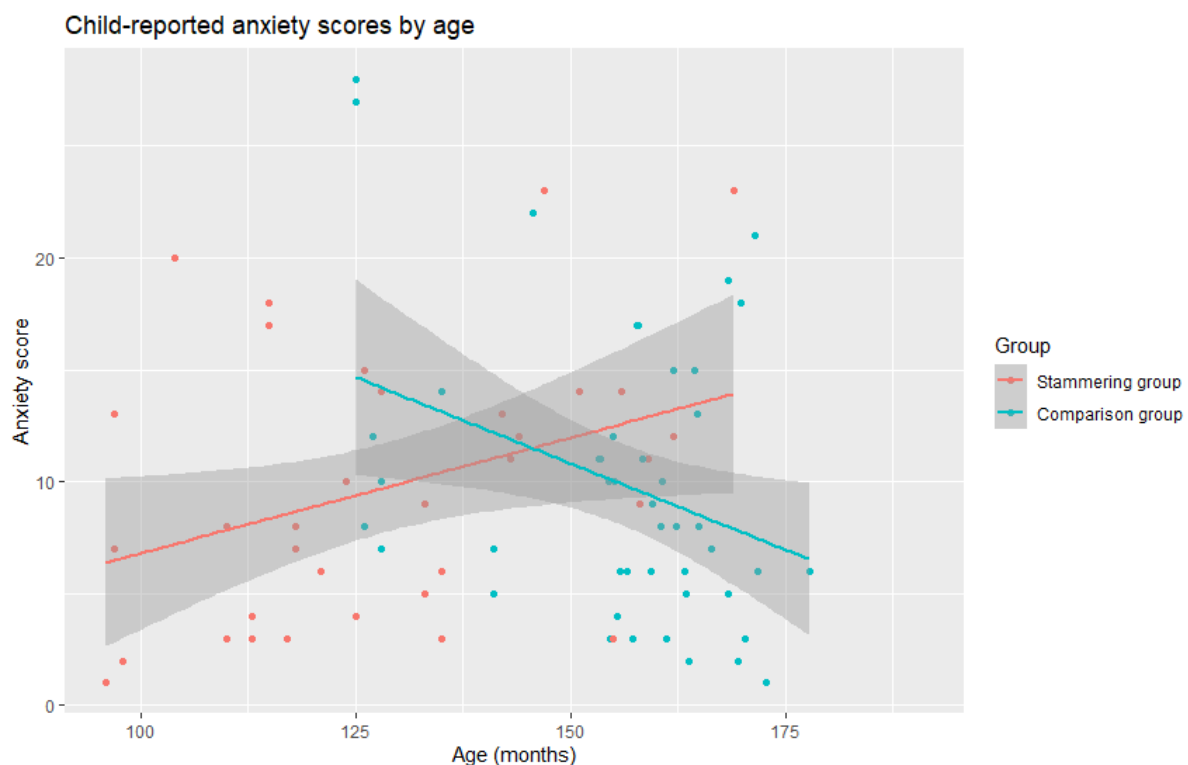


Figure 5: Child-reported anxiety scores by age in the stammering and non-stammering groups

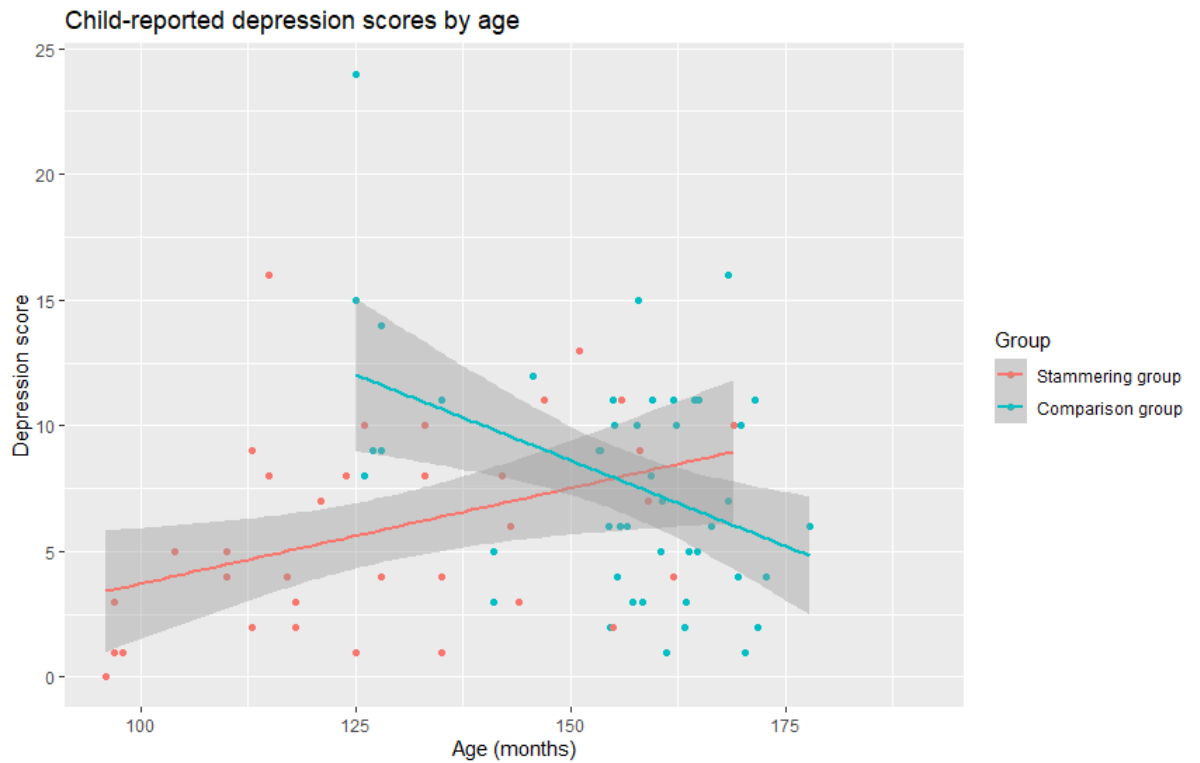


Figure 6: Child-reported depression scores by age in the stuttering and non-stuttering groups

Parent reported scores

Anxiety subscale scores

After controlling for age and SES, group was found to be a significant predictor of parent-reported anxiety scores ($b = -4.58$, $SE = 1.24$, $t = -3.68$, $p < .001$), with higher anxiety scores in the stuttering group. There was also a significant main effect of age, indicating that for every one month increase in child age, parent-reported anxiety scores increased by 0.08 points. This model was a significant fit to the data ($F(3, 80) = 5.85$, $p = .001$, adj. $R^2 = 0.149$), accounting for 14.9% of the variance in scores. I tested for an interaction effect between age and group in a third regression model but there was no significant interaction ($b = -0.08$, $SE = 0.06$, $t = -1.39$, $p = .17$).

Depression subscale scores

Parent-reported depression scores did not differ significantly between groups in the unadjusted analyses ($b = -0.96$, $t = -1.23$, $p = .22$), nor after accounting for age and SES, ($b = -1.69$, $t = -1.67$, $p = .10$). There was a significant effect

of age, suggesting that for every additional month in age there was a 0.05 point increase in scores. The adjusted model was found to be a significant fit to the data ($F(3, 80) = 3.65, p = .02, \text{adj. } R^2 = 0.0875$), accounting for 8.75% of the variance in scores. Again, two further models tested for interaction effects, but I found no interaction between age and group ($b = 0.02, SE = 0.05, t = 0.36, p = .72$) or between SES and group ($b = -0.0001, SE = 0.0001, t = -1.27, p = .21$) for parent-reported depression scores.

Table 7: Child-reported subscale scores for whole sample. Results of unadjusted and adjusted regression models

Anxiety subscale scores	Coefficient (b)	Standard Error (SE)	t-value	p-value
Model 1				
Group	0.19	1.40	0.14	.89
Model 2				
Age	0.15	0.05	2.75	.008
SES	-0.0001	0.00009	-1.36	.18
Group	43.7	11.9	3.67	<.001
Age*Group	-0.30	0.08	-3.69	<.001
Depression subscale scores				
Model 1				
Group	2.05	0.97	2.10	.04
Model 2				
Age	0.09	0.04	2.39	.02
SES	-0.0001	0.00006	-1.77	.08
Group	34.5	8.02	4.31	<.001
Age*Group	-0.22	0.05	-4.05	<.001

Note: Models based on child-reported anxiety and depression raw scores for the stuttering (n=35) and comparison (n=46) groups.

Table 8: Parent-reported subscale scores. Results of unadjusted and adjusted regression analyses.

Anxiety subscale scores	Coefficient (b)	Standard Error (SE)	t-value	p-value
Model 1				
Group	-2.11	0.97	-2.17	.03
Model 2				
Age	0.08	0.03	3.07	.003
SES	-0.00005	0.00007	-0.70	.49
Group	-4.58	1.24	-3.68	<.001
Depression subscale scores				
Model 1				
Group	-0.96	0.78	-1.23	.22
Model 2				
Age	0.05	0.02	2.27	.03
SES	-0.0001	0.00005	-2.12	.04
Group	-1.69	1.01	-1.67	.10

Note: Models based on parent-reported anxiety and depression raw scores for the stuttering (n=46) and comparison (n=46) groups.

Clinical vs subclinical scores

Mean T-scores for all subscales were below clinical threshold in both groups. Twelve participants obtained scores classified above threshold on at least one subscale. Of those in the comparison group that scored above clinical threshold on at least one of the subscales ($n = 7$), five reported a positive family history of adverse mental health, while two had previously accessed mental health services. Four reported speech and language concerns, while three of these participants had received diagnoses which may be associated with communication challenges (autism, speech & language disorder, sensory impairment). For five children in the stuttering group, T-scores exceeded the clinical cut-off on at least one subscale. Of these children, three were reported to have a positive family history of adverse mental health and one had previously accessed support for mental health.

Table 9 presents frequency of scoring 'above' or 'below' threshold on the child-reported and parent-reported anxiety and depression subscales. Scores were categorised as 'above threshold' (>70) or 'below threshold' (<65) for the purposes of analysis. Observation of the number of borderline

scores by group are reported for each subscale but were not included in analyses. The small sample size and low number of expected frequencies (<5) indicated that Fisher's Exact Test would be appropriate rather than Pearson's Chi-Square Test. There were no significant associations between group and scoring above clinical threshold on any subscale (Table 9).

Table 9: Frequency of scoring above or below clinical threshold based on T-scores in each group

Subscale	Group	Frequency Above N (%)	Frequency Below N (%)	Fisher's Exact Test
Self-reported Anxiety^a	Stuttering group	1 (3%)	33 (97%)	p = 1.0
	Comparison group	2 (4%)	43 (93%)	
Self-reported Depression^b	Stuttering group	0	34 (100%)	p = 1.0
	Comparison group	1 (2%)	42 (91%)	
Parent-reported Anxiety^c	Stuttering group	5 (11%)	39 (87%)	p = 0.27
	Comparison group	2 (4%)	43 (93%)	
Parent-reported Depression^d	Stuttering group	3 (6.6%)	38 (84%)	p = 1.0
	Comparison group	3 (6.5%)	42 (91%)	

Note. For child-reported scores: stuttering sample (n=34), comparison group (n=46).

For parent-reported scores: stuttering sample (n = 45), comparison group (n=46).

^a Data unavailable for 12 participants in the stuttering group. One participant in the comparison group obtained a borderline score and were not included in the analysis.

^b Data unavailable for 12 participants in the stuttering group. Three participants in the comparison group obtained a borderline score and were not included in the analysis.

^c Data unavailable for 1 participant in the stuttering group. One participant in each group obtained a borderline score and were not included in the analysis.

^d Data unavailable for 1 participant in the stuttering group. Four participants in the stuttering group and one in the comparison group obtained a borderline score and were not included in the analysis

Association between anxiety and depression scores

Pearson Product Moment Correlation Coefficients were estimated to address the hypothesis regarding the association between anxiety and depression scores in the stuttering group. A positive, moderate correlation was found between child-reported anxiety and depression scores ($r(33) = 0.59$, 95% CI[0.32, 0.77], $p < .001$). There was a strong, positive correlation between parent-reported anxiety and depression scores ($r(44) = 0.70$, 95% CI[0.52, 0.83], $p < .001$). Similarly, in the comparison group, anxiety and depression symptoms were positively correlated in child-reported ($r(44) = 0.82$, 95% CI[0.70, 0.90], $p < .001$) and parent-reported ($r(44) = 0.63$, 95% CI[0.41, 0.78], $p < .001$) scores.

Differences in scores by respondent

The results of Welch two sample t -tests indicated parent- and child-reported scores did not differ significantly for anxiety ($t(67.1) = 1.74$, $p = .35$, $d = 0.39$) or depression ($t(69.2) = 0.87$, $p = 1.0$, $d = 0.20$) subscales in the stuttering group. In contrast, children reported significantly more symptoms of anxiety ($t(76.9) = 3.97$, $p < .001$, $d = 0.83$) and depression ($t(87.7) = 4.17$, $p < .001$, $d = 0.87$) than their parents in the comparison group.

Exploratory Analysis: Inter-rater reliability

In addition to the original hypothesis concerning the extent to which symptoms scores differed by respondent, exploratory analyses were conducted to investigate levels of respondent agreement. In order to assess inter-rater reliability between respondents, Intraclass Correlation Coefficients (ICC) were calculated for the two groups. One-way random effects models estimated absolute agreement based on an average rating between parent and child scores in each group.

In the stuttering group ($n = 35$), ICC estimates indicated good agreement between parent and self-reported anxiety (ICC = 0.71, 95% CI[0.43, 0.85], $p < .001$) and depression (ICC = 0.85, 95% CI[0.71, 0.92], $p < .001$) scores.

However, in the comparison group ($n = 46$), agreement between parent- and self-reported anxiety ($ICC = 0.17$, $95\%CI[-0.50, 0.54]$, $p=.27$) and depression ($ICC = -0.14$, $95\%CI [-1.06, 0.37]$, $p = .67$) symptoms was poor.

Discussion

The primary objective of this study was to evaluate whether children who do and do not stutter differ significantly in self- and parent-reported symptoms of anxiety and depression. The relationship between group and age differed for both child-reported anxiety and depression symptoms, with increasing age associated with higher scores in the stuttering group and with lower scores in the comparison group. This interaction was not observed in parent-reported scores. Instead, parent-reported anxiety scores were significantly higher for children who stutter compared to children who do not stutter when age and SES were controlled for. However, there were no significant differences in parent-reported depression scores after accounting for age and SES.

The observed relationship between increasing age and greater anxiety symptoms in children who stutter aligns with previous hypotheses (e.g. Smith et al., 2014). However, these results are not conclusive; future research is needed to replicate the present findings in larger samples using longitudinal designs to track symptoms over time. Parent-reported scores indicate that children who stutter experience higher anxiety symptoms, but not depression, relative to children who do not stutter. Nevertheless, for most participants, scores did not reach clinical threshold and, as a group, children who stutter were not more likely to present with clinical level symptom scores.

Association between anxiety and depression symptoms

Anxiety and depression scores were positively correlated in both groups, suggesting that in the case of parent and child report, higher anxiety symptoms are associated with higher depression symptoms. High co-occurrence of anxiety and depression in young people may relate to the

overlap of symptoms, presence of shared risk factors and evidence that the constructs of anxiety and depression are not totally distinct, instead sharing a common component while also having distinguishing features (see Angold, Costello & Erkanli, 1999; Axelson & Birmaher, 2001; Seligman & Ollendick, 1998).

Despite commonly co-occurring in the general population, there has been comparatively less research into depression symptoms in this population. This is an important gap in the literature, especially as depression in adolescence is associated with later adverse outcomes (see Thapar et al., 2012). The present findings therefore reinforce the need for further investigation into the development of both anxiety *and* depression in young people who stutter using longitudinal research designs. This would serve to enhance our understanding of lifetime risk for depression, potentially supporting timely identification and clinical management.

Discrepancies in anxiety and depression symptom reporting

In the present study, there was good agreement between parent- and child-reported symptom scores in the stuttering group. This is in contrast to the discrepancies in respondent reporting documented in the literature (De Los Reyes and Kazdin, 2005). Perhaps more consistent with previous reports of respondent discrepancies in mental health, intra-class correlation coefficients indicated poor agreement between parent- and self-reported anxiety and depression symptom scores in the comparison group, with children reporting more symptoms on average relative to parent reports.

There are likely to be a number of child, parent and family factors that moderate these discrepancies in symptom ratings, such as the child's age, social desirability, the level of distress the child experiences, and parental depression or anxiety (for a review see De Los Reyes & Kazdin, 2005). Given the small sample size, it is difficult to determine why parent and child scores substantially differed, though in this study responses may reflect the circumstances or recent experiences of families at the time of survey

completion. For example, in the SCALES study, children completed the RCADS-25 during the online session with the researcher, while parents were asked to complete it online or post it back to the research group during the testing phase. Conversely, for the stuttering group, the parent and child versions of the RCADS-25 were completed through the same online questionnaire. While participants were encouraged to allow children to complete the RCADS-25 independently, it is possible that parental presence may have affected children's responses. Children may have provided more favourable responses as they were aware their parents could see their answers, while some children may have needed support completing the survey. Previous studies have also suggested that children and adolescents who stutter may conceal their true levels of anxiety when completing self-report measures in an effort to present themselves in a more positive light (Gunn et al., 2014; Messenger et al., 2015). Finally, all children in the stuttering group had accessed intervention and half reported a positive family history of stuttering. It may be that parents and children were more familiar with talking about the emotional consequences of stuttering due to intervention and parental experience of the potential challenges associated with stuttering. This may have offered more opportunities to share experiences and inform perceptions about the impact of stuttering in this group. Regrettably, the data does not permit further exploratory analysis of this hypothesis as we do not have details of the nature of the intervention received.

Factors contributing to symptom scores

The present study contributes further evidence of the heterogeneity in internalising symptom reporting amongst children who stutter. Some previous studies have found no significant differences in self-reported anxiety in school-aged children who do and do not stutter (e.g. Andrews & Harris, 1964; Craig & Hancock, 1996; Rocha, Yaruss & Rato, 2019), while others have reported significantly higher anxiety symptoms in school-aged children and adolescents who stutter relative to non-stuttering peers (e.g. Blood et al., 2007; Blood & Blood, 2007; Mulcahy et al., 2008; Iverach et al., 2016;

Veerabhadrapa, Vanryckeghem & Maruthy, 2021). The null findings for the parent-reported depression subscale are consistent with much of the literature evaluating depression symptoms in school-aged children and adolescents who stutter (see Bernard, Hofslundsengen & Norbury, 2022; Livingstone-Pountney & Mitrevski, 2019). However, the mean age of the present sample may have meant that it was too early to detect meaningful differences between groups, especially as prevalence of depression increases in late adolescence (Thapar et al., 2012).

A number of child and family factors may have contributed to the variability in subscale scores in the stuttering group. First, all the children who stutter in the current study had accessed Speech and Language Therapy, which may have reduced anxiety (Craig, 1994; Menzies et al., 1999). Speech and language therapy for stuttering may provide children with direct support for improving speech fluency, which may have improved self-esteem and confidence, and/or promote strategies to manage the psychosocial elements associated with stuttering, such as negative attitudes and anxieties around speaking.

Secondly, as this study relied on two population samples, I did not exclude children with other communication difficulties. The proportion of the stuttering group with co-existing diagnoses is unknown. However, almost one quarter of children in the comparison group reported a clinical diagnosis, and half of those scoring above clinical threshold in the comparison group had an existing clinical diagnosis. There were insufficient data to perform further sub-group analyses. Nevertheless, given the association between increased risk for anxiety and other neurodevelopmental conditions, such as developmental language disorder and autism (Kim et al., 2000; Yew & O’Kearney, 2013), these additional difficulties may have contributed to symptom reporting. Some studies have reported higher anxiety symptoms in children who stutter with co-occurring disorders compared to children who stutter without co-occurring disorders (Alm, 2014; Blood et al., 2007; Smith et al., 2017). Future studies could explore whether elevated anxiety in children

and adolescents who stutter is associated with stuttering per se or presence of co-existing developmental difficulties.

Thirdly, more children in the stuttering group reported a positive family history of mental health difficulties relative to those in the comparison group, although the proportion of those who accessed professional support for mental health was similar across groups. Positive family history of anxiety and depression is associated with increased risk for internalising problems in children (Pine & Klein, 2015; Thapar et al., 2012). This may have contributed to the significantly higher parent-reported anxiety scores in the stuttering group. Nonetheless, relative to the number of children who reported a positive family history of mental health issues, very few children scored in the clinical range. Not all children considered at-risk of developing mental health difficulties will go on to present with internalising problems, highlighting the role of resilience and interaction of risk and protective mechanisms (Rutter, 1987; Thapar et al., 2012).

The RCADS-25 may not be a sufficiently sensitive measure for evaluating the psychosocial experiences of children who stutter. Jones et al. (2021) concluded that there were insufficient measures for adequately assessing the psychological impact of stuttering in children. To the best of my knowledge at the time of writing, this is the first study to use the RCADS-25 to evaluate anxiety and depression in children who stutter in a community sample. While the RCADS-25 has good psychometric properties and is widely used in clinical and research contexts (Ebesutani et al., 2012; Piqueras et al., 2017), the questions are not specific to the experience of stuttering. It may be that measures designed to align more closely with the experience of stuttering, and that purport to evaluate social anxiety and speech-related anxiety may have resulted in different findings. For instance, Veerabhadrapa et al. (2021) recently reported significantly higher scores on the Speech Situation Checklist in children who stutter (aged 7 – 14 years) compared to children who do not stutter.

Finally, the symptoms reported by both groups may have reflected the novel coronavirus context and associated disruption to everyday life. The impact of the COVID-19 pandemic on children who stutter is as yet unclear.

Nevertheless, one hypothesis for the fact children who stutter did not report significantly elevated internalising symptoms relates to reduced exposure to some of the environmental factors, such as bullying and negative peer reactions, during the period of school closures. Negative reactions from peers and bullying victimisation have been associated with negative psychosocial outcomes, such as low self-esteem and elevated anxiety, in the stuttering (Blood & Blood, 2004; Blood & Blood, 2007; Cook & Howell, 2014) and non-stuttering populations (Lereya, Copeland, Costello & Wolke, 2015; Schoeler, Duncan, Cecil, Ploubidis, Pingault, 2018). The reliance on online teaching has meant that, for some children at least, there may have been less pressure to vocally contribute in class and alternative options made available to participate in lessons, such as use of the chat function on platforms like Zoom or Microsoft Teams. On the other hand, some of the rules and restrictions associated with the pandemic may have created greater communication challenges, such as wearing face masks that obscure the speakers' face and consequently hide silent blocking behaviours resulting in interruptions by listeners.

Limitations

Data were collected predominantly during the global coronavirus pandemic which affected recruitment and altered everyday experiences in ways that have impacted well-being. Results must be interpreted with caution and cannot be generalised to the experiences of children who stutter pre-pandemic. As this was a cross-sectional design and pre-pandemic scores were not available, it is not possible to speculate as to whether children responded any differently than they would have done outside of the pandemic. Nevertheless, national lockdowns are likely to have had a substantial (and predominantly negative) impact on the mental health of children and young people as evidenced by early findings (Cowie & Myers, 2021; Loades et al., 2020).

While the decision to host the questionnaire online was intended to improve the representativeness of the stuttering sample, families with restricted access to a computer or internet connection are less likely to take part. Additionally, while I did not recruit from clinic waiting lists, families who engage with stuttering organisations and charities may differ to families who do not, introducing selection bias. For instance, parents who are particularly concerned about their child's stutter may be more likely to seek support, while clinical services may highlight these organisations to families during the course of intervention.

I had intended to recruit a minimum of 100 participants. Despite the questionnaire remaining open for over a year, data for only 46 participants who stutter contributed to the findings. This sample size reduces statistical power. Furthermore, the small sample size meant that it was not possible to conduct further planned analyses that may have contributed to the interpretation and our understanding of these findings through consideration of possible moderating factors. Nonetheless, confidence intervals have been reported, in preference to post-hoc power analyses, to aid interpretation (Heckman, Davis & Crowson, 2022). Additionally, the pandemic prompted a change to the pre-registered protocol whereby scores could no longer be compared with pre-pandemic population norms. Instead it was necessary to compare the stuttering group to a non-stuttering comparison group. As the comparison group had not been recruited specifically for this study, differences existed both in administration of the questionnaire and the demographic characteristics of the groups that otherwise could have been controlled for. This may have resulted in practice effects in the non-stuttering group, while group differences may have influenced symptom reporting. Consequently, it is difficult to draw firm conclusions about the varying effect of age on symptom reporting given the differences in mean age and age range in either group.

Conclusion

The present findings suggest that stuttering may be associated with increasing symptoms of anxiety, and possibly depression, as children get older and move into adolescence. Despite the novel circumstances in which this study was conducted, the findings nonetheless underline the heterogeneity in this clinical population with regards to experience of anxiety and depression symptoms. Future research should prioritise longitudinal, population studies that account for known risk factors associated with adverse mental health and track symptoms into adulthood, when increased risk for anxiety has been more consistently reported. Further examination of depression symptoms in this population is also warranted given the moderate correlation between anxiety and depression observed in this study and the wider literature. It is important to improve our understanding of the mechanisms underlying variable anxiety outcomes reported for children and adolescents, as this could enhance clinicians' awareness of risk and resilience in this population.

Chapter 4

Factors associated with anxiety and depression symptoms in children who stutter

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Abstract

Purpose. Children and adolescents who stutter may be at risk of elevated anxiety and depression symptoms, although studies have indicated variability in reported internalising symptoms in this population. This study considers the association between anxiety and depression symptoms and stuttering, as well as child, family, and contextual factors that may affect this association.

Method. Thirty-five school-aged children who stutter completed the Revised Children's Anxiety & Depression Scale-short version (RCADS-25).

Regression models were fitted to the data to examine the association between anxiety and depression symptoms with bullying, stuttering severity, family history of adverse mental health, and age in children who stutter.

Results. Family history of adverse mental health was found to significantly predict anxiety and depression scores. Age also predicted depression scores, with older children reporting higher scores.

Conclusion. Family history of adverse mental health is associated with higher self-reported internalising symptoms in children who stutter. The interaction between child, family, and contextual factors may change with age, and this requires further exploration in larger, longitudinal studies. The association between bullying and anxiety scores indicates the importance of anti-bullying initiatives in promoting psychosocial development in school-aged children who stutter. This study also highlights the contribution of known risk factors for mental health, such as family history, to variability in symptom reporting.

Introduction

Developmental stuttering is a complex, neurodevelopmental condition characterised by behavioural, affective, and cognitive features (Bloodstein, et al., 2021). Behavioural features include speech characteristics that result in disruptions in the rhythm and timing of speech, such as repetitions, prolongations, and blocks of airflow, as well as secondary behaviours, such as the head nods, blinking, and muscle tension observed in some individuals who stutter. These disruptions in spoken fluency may be accompanied by emotional responses (affective features), such as fear of speaking (Bloodstein et al., 2021). Additionally, cognitive factors may develop over time and include attitudes and beliefs about stuttering and communicating, such as anticipation of stuttering and avoidance of words and situations (Bloodstein et al., 2021).

Many individuals who stutter report negative attitudes from peers, bullying victimisation, and teasing related to their speech (Blood et al., 2011; Blood & Blood, 2007; Davis et al., 2002). Stuttering has also been associated with low self-perceived communication competence, heightened communication apprehension and fear of negative evaluation in adolescence (Erickson & Block, 2013; Mulcahy et al., 2008; Blood et al., 2001). These negative experiences may put children and young people who stutter at greater risk of adverse mental health outcomes, particularly anxiety, relative to non-stuttering peers (Smith et al., 2014).

Internalising conditions refer to those characterised by disordered mood or emotion, such as anxiety and depression (Kovacs & Devlin, 1998; Liu et al., 2011). Internalising problems in childhood are associated with adverse educational, social, and quality of life outcomes (Liu et al, 2011; Stevanovic, 2013). Anxiety and depression are among the most common mental health disorders that affect children and young people, and commonly co-occur in adolescents and adults (Brady & Kendall, 1992; Lewinsohn et al., 1997). Anxiety refers to a negative emotional state involving anticipation of threat, which although a natural fight or flight response, becomes pathological when

it interferes with everyday functioning (Essau, 2007). Depression is characterised by excessive sadness and loss of interest in otherwise enjoyable activities (Liu et al., 2011). While the onset of anxiety may occur in childhood, depression typically develops during mid to late adolescence (Huberty, 2012). Several social, familial, and psychological factors are associated with increased risk for anxiety and mood disorders among children and adolescents, including female sex, economic deprivation, parental history of either condition, childhood adversity and stressful life events (Hyland et al., 2016; Kovacs & Devlin, 1998). Experiencing anxiety and depression in adolescence also increases the likelihood for recurrence in adulthood (Kovacs & Devlin, 1998), further underscoring the importance of early identification and appropriate support for childhood mental health.

Community studies have indicated that stuttering in adolescence is associated with higher levels of psychological distress and holding lower status jobs in adulthood (McAllister et al., 2013; McAllister et al., 2012). It has been suggested that the difficulties with communication, negative peer reactions, and bullying experienced in social contexts may increase the risk for anxiety amongst children who stutter (Briley et al., 2019; Smith et al., 2014). Improving our understanding of factors that may be involved in any association between internalising problems and stuttering would serve to inform clinical management of this population. If children who stutter are at elevated risk of internalising problems relative to children who do not stutter, there is a need to focus on how clinicians work with this population to offset the adverse consequences of poor mental health.

Stuttering and mental health

Stuttering can be associated with negative attitudes and emotions, and adults who stutter experience increased risk for anxiety and social anxiety disorder compared to the non-stuttering population (Craig & Tran, 2014; Iverach & Rapee, 2014). It has been proposed that maintenance of social anxiety disorder in adults who stutter may be related to several interrelated risk factors, such as fear of negative evaluation, attentional biases, negative

social-evaluative cognition and safety behaviours, such as avoiding socially threatening situations and difficult words (Iverach et al., 2017a).

However, findings regarding anxiety in children who stutter have been more mixed. A recent meta-analysis indicated that some children and adolescents who stutter are at risk of elevated anxiety symptoms relative to non-stuttering peers ($g = 0.42$, 95% CI [0.10, 0.74]), although findings were variable across studies (Bernard et al., 2022). Previous reviews of the literature have cited several factors that may contribute to these inconsistent findings, such as small sample sizes, under-powered analyses, selection bias, use of anxiety measures that are insufficiently sensitive to the experiences of individuals who stutter, and limited control of potential confounding variables, including co-occurring disorders and previous mental health intervention (Bernard et al., 2022; Menzies et al., 1999; Smith et al., 2014). Although the approximate age of onset for anxiety in children who stutter is unclear, Smith et al. (2014) suggested that anxiety levels may increase as children reach adolescence and young adulthood until they exceed normal limits.

There have been fewer studies to date focusing on depression symptoms in the stuttering population, despite the comorbidity between anxiety and depression in the typical population (Brady & Kendall, 1992; Lewinsohn et al., 1997). There appears to be little evidence that children and adolescents who stutter (< 18 years) report elevated depression symptoms (Bernard et al., 2022; Livingstone-Pountney & Mitrevski, 2019). However, some studies have found a significant relationship between stuttering and depression in adults (see Livingstone-Pountney & Mitrevski, 2019), while higher anxiety scores have been found to significantly predict higher depression symptoms in adolescents seeking treatment for stuttering (Iverach et al., 2017b).

Contextual factors: Bullying and mental health

Bullying or victimisation can be defined as repeated exposure to negative actions by one or more peers, and typically implies an imbalance of strength or power (Olweus, 1993). Negative actions are characteristic of aggressive

behaviours in which an individual intends to inflict injury or discomfort; these may be verbal, for example teasing and name calling, or physical, for instance kicking and punching. Similarly, a distinction can be drawn between direct bullying, in which the victim is openly attacked, and indirect bullying, which may involve efforts to socially exclude an individual (Olweus, 1993).

Bullying is associated with a number of educational and health consequences during childhood, adolescence, and adulthood, which are determined by the frequency and type of bullying, whether the individual is the perpetrator, victim or both, and the point at which these consequences are observed (Armitage, 2021). Meta-analyses of cross-sectional and longitudinal studies indicate a strong association between bullying victimisation in childhood and internalising problems, particularly depression and anxiety (Moore et al., 2017; Reijntjes, et al., 2010). Bullying experienced in childhood and adolescence has also been associated with increased risk for depression and anxiety in adulthood (Lereya et al., 2015; Sigurdson et al., 2015). On the other hand, Schoeler et al. (2018) performed a meta-analysis of quasi-experimental studies ($k = 16$) to examine the consequences of bullying victimisation and found that the adverse effects on mental health reduced in the longer term. They concluded that bullying may be causally associated with adverse mental health outcomes in the short term, especially anxiety and depression, but that the reduction in these effects over time highlights the potential for resilience (Schoeler, et al., 2018).

Peer reactions and bullying in childhood stuttering

Children and adolescents who stutter appear to be at increased risk of negative peer reactions, peer rejection, and bullying (Blood & Blood, 2004; Blood & Blood, 2007; Davis et al., 2002). Negative peer reactions may be associated with increasing age and stuttering severity (Bloodstein et al., 2021). Blood et al. (2011) reported that 44% of the adolescents who stutter (13 – 18 years) experienced bullying victimisation, compared to 9% of the non-stuttering group. A higher proportion of adults who stutter have also retrospectively reported being bullied in primary and secondary school

(30.6% and 27.8% respectively) compared with 13.9% of the non-stuttering group (Blood & Blood, 2016). Using the Teasing/Bullying Questionnaire for Children who Stutter (TBQ-CS; Langevin et al., 1998), studies have reported bullying victimisation and teasing in 53% to 59% of children and adolescents who stutter (Erickson & Block, 2013; Langevin et al., 1998).

Similar to the general population, bullying victimisation has been associated with poorer psychosocial and quality of life outcomes in adolescents who stutter. Studies have reported that bullying victimisation correlates with lower self-esteem, poorer communication competence, and higher anxiety scores (Blood & Blood, 2004; Blood & Blood, 2007; Blood et al., 2011; Cook & Howell, 2014). The increased rates of bullying experienced by children and adolescents who stutter may therefore increase risk for anxiety and depression in this clinical population.

Child factors: stuttering severity

Stuttering severity could be considered a risk factor for elevated anxiety as more frequent disruptions in fluency may result in negative reactions from peers. For instance, when listening to school-aged children who stutter, positive reactions from peers decrease as stuttering severity increases (Panico et al., 2015).

Negative communication attitudes and poor self-perceived communication competence have also been observed in children and adolescents who stutter relative to peers who do not stutter, and these negative speech-related attitudes have been found to increase with age but not differ by sex (Blood et al., 2001; Guttormsen et al., 2015). Blood et al. (2001) found that stuttering severity was significantly associated with poorer self-perceived communication competence and higher communication apprehension in adolescents who stutter. Studies with children who stutter have also shown that negative communication attitudes increase with stuttering severity (Kawai et al., 2012; Vanryckeghem & Brutten, 1996). Negative communication attitudes have been found to mediate the relationship

between stuttering severity and self-esteem in adolescents who stutter (Adriaenssens et al., 2015). Low self-esteem is associated with internalising disorders, particularly depression, in children and young people (Keane & Loades, 2017). Therefore, the severity of one's stutter may affect the way an individual perceives their communicative ability, which could have implications for self-reported emotional well-being, such as anxiety or depression.

However, evidence that stuttering severity is associated with anxiety levels has been mixed. Although they found stuttering severity to be significantly associated with negative communication attitudes, Miller and Watson (1992) reported no significant association between self-rated stuttering severity and anxiety or depression in adults who stutter. Craig et al. (2003) found that adults with more severe stuttering did not report significantly higher trait anxiety than those with less severe stuttering, as measured by per cent syllables stuttered (%SS). On the other hand, Ezrati-Vinacour and Levin (2004) found that stuttering severity was related to state anxiety, which is situation dependent. This led them to conclude that "it is not the mere stuttering that perpetuates the state anxiety but the severity of the stuttering accounts for the extent to experience anxiety in social communication" (Ezrati-Vinacour & Levin, 2004, p143). Several studies have found no significant correlation between stuttering severity and anxiety symptom scores in children and adolescents (see Bernard et al., 2022), though others have (Iverach et al., 2017b; Veerabhadrappe et al., 2021).

Family factors: history of mental health and stuttering

A family history of mental health conditions, particularly parental anxiety and depression, are acknowledged risk factors for child anxiety and depression (Hyland et al., 2016; Thapar et al., 2012). Examination of social, psychological, and family risk factors in anxiety and mood disorders highlights the cumulative effect of a number of risk factors in development of internalising problems (Hyland et al., 2016). As well as child and contextual factors that may increase risk for elevated symptoms in stuttering, family

history of mental health issues may also be contributing to the variability in symptom reporting across studies. It was therefore hypothesised that family history of adverse mental health would have a significant effect on anxiety and depression scores in the present sample.

Stuttering is a disorder with high heritability (Frigerio-Domingues & Drayna, 2017). It was anticipated that a substantial proportion of respondents would report a positive family history of stuttering in the sample. Positive family history of stuttering may influence parents' perceptions of the impact of stuttering on their children (Rocha et al., 2020), which could affect the way parents and children interact. Family experience of stuttering may also result in increased understanding of the condition, the challenges it can pose and the broader experience of living with a stutter. This may mean that families can draw on their own experiences and strategies to support their children. I hypothesise that increased exposure to stuttering through close family members will facilitate greater understanding and support, which may help to offset adverse mental health symptoms.

The present study

This study aimed to investigate the factors that affect associations between anxiety and depression symptoms and stuttering in children. In this study, I consider the association between self-reported anxiety and depression symptoms and child, family, and contextual factors identified from the literature that may predict anxiety and depression symptoms in this population. The Revised Children's Anxiety and Depression Scale-short version (RCADS-25) was used to measure self-reported anxiety and depression symptoms in a cohort of children who stutter in the UK. Should RCADS-25 scores, bullying and stuttering severity ratings correlate, I intend to explore whether bullying victimisation mediates the relationship between stuttering severity and RCADS-25 subscale score.

Because a smaller sample than anticipated was recruited, I focused on only three of the pre-registered hypotheses (<https://osf.io/cr392/>) for this study, which are as follows:

- There will be an association between family history of mental health conditions and RCADS-25 scores.
- There will be a negative association between family history of stuttering and RCADS-25 scores.
- Self-reported experience of bullying mediates the relationship between stuttering severity and RCADS-25 scores.

Method

This study was pre-registered on the Open Science Framework in October 2019 and amended in March 2021 in response to the COVID-19 pandemic prior to data analysis (<https://osf.io/4fgex>). Ethical approval was granted by University College London (UCL) Research Ethics Committee (15535/001). Parental consent and child assent were obtained at the start of the questionnaire.

Participants

(i) *Recruitment*

Details about the study were made available on the website of a UK stammering charity and advertised to families engaged in the work of that charity. Additionally, information about the study was advertised through other organisations in the UK that work with families of children who stutter and speech and language therapists, online support groups and via social media platforms. Study leaflets were distributed to mainstream primary and secondary schools in London. Participants were recruited to this study through volunteer self-selection; individuals who registered interest in the study were sent a link to the questionnaire.

Inclusion criteria to take part in the questionnaire were as follows: (i) child has a developmental stutter; (ii) child is aged between 8 and 13 years; (iii)

child lives in the UK. Five participants who completed the questionnaire fell outside this age range. As the Revised Children's Anxiety and Depression Scale (RCADS-25) is normed on children aged 8 – 18 years, data for children under eight were removed ($n = 3$). One participant aged 14 years was retained in the analyses. Demographic data (age, sex, SES) were unavailable for one participant but complete data for all outcome and predictor variables was available for this individual and therefore their data were retained in the analyses. Participants completed the questionnaire between February 2020 and August 2021.

(ii) *Sample characteristics.*

In total, 46 families took part in the questionnaire, however only 35 children completed the child version of the RCADS-25 and therefore the final sample for this study included 35 children and adolescents. Background information, such as family history, access to intervention, and confirmation of stuttering, was available for all participants ($n = 35$). Complete demographic data (sex, age, socio-economic status) were available for 30 participants: four participants had no SES data and one participant reported no information about sex, age or SES. I imputed the mean sample age for the sole participant with missing age data.

Socio-economic status (SES) was measured using the Income Deprivation Affecting Children Index (IDACI), which is a supplementary index of the English Indices of Deprivation (IoD) and measures the proportion of children (0-15 years) living in income deprived families (Ministry of Housing, Communities & Local Government, 2019). The IoD ranks all neighbourhoods in England from 1 (most deprived area) to 32,844 (least deprived area). For those living outside of England, it was not possible to calculate the IDACI rank as a UK-wide version of the IoD is unavailable. SES data were missing for five participants in total.

The sample comprised nine female and 25 male participants (one undisclosed), with a mean age of 10 years ($M = 128$ months, $SD = 20.4$

months). All participants spoke English as their first language, and six participants reported speaking an additional language. The IDACI ranks for this sample ranged from 2,569 to 32,200 ($M = 17,868$, $SD = 8,404$). The reported age of stuttering onset ranged from two to seven years old ($M = 3.69$, $SD = 1.48$). All participants reported that they had accessed speech and language therapy and 31/35 had received a diagnosis of stuttering from a speech and language therapist. Half of participants reported a family history of stuttering ($n = 18$) and nine of these participants did not have a family history of mental health issues. Of the participants with a positive family history of mental health difficulties ($n = 18$), nine did not have a family history of stuttering. A quarter of participants ($n = 9$) reported a positive family history of both mental health and stuttering. Only three respondents reported that their child had accessed support for mental health previously. Sample characteristics can be found in Table 10.

Online questionnaire

The authors involved individuals with experience of stuttering in the development of the questionnaire by piloting it with the youth panel of a stuttering charity, composed of young people who stutter (aged 16 – 25 years) who advocate for greater public awareness about stuttering, and the charity's board of trustees. Responses collected as part of the pilot testing of the questionnaire were not included in the final sample. The questionnaire comprised two sections, one for parents and one for children who stutter. Parents completed the background questionnaire and the RCADS-25-P (Ebesutani et al., 2017). The children's section of the questionnaire involved completion of the RCAD-25-C (Ebesutani et al., 2012), Illinois Bullying Scale (IBS; Espelage & Holt, 2001) and self-perceived stuttering severity rating scale. The questionnaire asked that children complete their section independently. Families had up to 60 days to complete the questionnaire and did not have to complete it in one sitting.

Outcome variable

Revised Children's Anxiety & Depression Scale-short version.

The Revised Children's Anxiety & Depression Scale (Chorpita et al., 2000) is a self-report measure that assesses the frequency of anxiety and low mood symptoms relating to DSM-IV symptom criteria in children and adolescents between 8 and 18 years of age. It is one of the Child Outcome Research Consortium (CORC) measures.

A shortened version of the RCADS (RCADS-25; Ebesutani et al., 2012) is available, which provides three scores: 'Total Anxiety', 'Total Depression', and 'Total Anxiety & Depression'. The RCADS-25 is based on the full 47-item RCADS, and retains the 10-item Depression Scale from the full version. However, the Anxiety Scale comprises 15-items and these items are based on the five subtypes of anxiety measured in the full RCADS. Given that I was interested in broad anxiety symptoms and wanted to optimise questionnaire completion, the RCADS-25 was used rather than the 47-item RCADS.

There are parent and child versions of the RCADS-25 (RCADS-25-C, Ebesutani et al., 2012; RCADS-25-P, Ebesutani et al., 2017), which allow for symptom reporting to be obtained from the child (self-report) and their parent. For the purposes of the present analyses, data obtained from the child version of the RCADS-25 (RCADS-25-C) are reported. Similarly to the RCADS, the RCADS-25-C illustrates good discriminatory validity for anxiety and depression. Both the Depression Scale and Anxiety Scale have good reliability in clinical ($\alpha = 0.80$, $\alpha = 0.96$ respectively) and school ($\alpha = 0.79$, $\alpha = 0.94$ respectively) samples (Ebesutani et al., 2012; Ebesutani et al., 2017). Normative data are provided to calculate T-scores that indicate whether symptom scores are in the normal, borderline or clinical range (see Ebesutani et al., 2012).

Predictor variables

Predictor variables were extracted mainly from the background questionnaire completed by parents. These included factors associated with internalising

symptoms, such as sex, age and family history of mental health. Stuttering severity and bullying victimisation measures were based on child-report.

Illinois Bullying Scale

The Illinois Bullying Scale (IBS; Espelage & Holt, 2001) is an 18-item self-report questionnaire that assesses frequency of peer victimisation, bullying behaviour, and fighting in children and young people (8 – 18 years). It is one of the measures included in the Compendium of Assessment Tools for measuring bullying victimisation, perpetration, and bystander experiences (Hamburger et al., 2011). The IBS comprises three subscales: Bully, Fight, and Victim. Subscale scores are calculated by summing the scores for subscale items. I was particularly interested in responses to the Victim Subscale as this measures the frequency with which an individual experiences bullying victimisation. Participants are presented with a series of statements and asked to indicate how often they experienced the activity in the last 30 days. The victim subscale comprises the following four statements: *'other students picked on me'*, *'other students made fun of me'*, *'other students called me names'* and *'I got hit or pushed by other students'*. Scoring corresponds to the choice of response: 'never' (0), '1 or 2 times' (1), '3 or 4 times' (2), '5 or 6 times' (3) or '7 or 8 times' (4). Scores on the Victim Subscale range from 0 – 16, with higher scores indicating more victimisation. Norms are unavailable for this scale and instead scores indicate frequency of bullying victimisation. Good reliability ($\alpha = 0.88$) has been reported for this scale (Espelage & Holt, 2001).

Self-perceived stuttering severity

Stuttering severity can be measured in terms of overt surface features, such as stuttering frequency, tension and duration, as well as more subtle features, including word avoidance, escape behaviours, and affective responses (Manning & DiLollo, 2018). Researchers and clinicians typically assess stuttering frequency by estimating the percentage of stuttered syllables (%SS) or words (%SW). However, self-rating scales of stuttering severity can offer insight into the individuals' perception of their stutter and

can be useful in initial clinical assessment (Manning & DiLollo, 2018). In this study, children were asked to self-rate the severity of stuttering on a Likert-style scale (1 = not very severe/bumpy, 10 = very severe/bumpy). Self-rating scales have been found to correlate well with clinician ratings in adults who stutter (O'Brian et al., 2004), and have been used in previous studies of anxiety in children who stutter (Gunn et al., 2014; Iverach et al., 2017b; Messenger et al., 2015). I chose to include a 9-point self-rating scale as I was interested in the association between a number of self-report measures and because it offered a simple and time-efficient way of obtaining a measure of stuttering severity from the child's perspective.

Family history of mental health

Families were asked whether there was a history of mental health conditions in their family as part of the background section of the online questionnaire. Families responded 'yes', 'no', 'not sure' or 'prefer not to say' and were asked to include the types of difficulties experienced, for example anxiety, depression, bipolar disorder. These data were coded as follows for the purposes of analyses: no = 0 (negative family history), yes = 1 (positive family history), not sure/prefer not to say = 2.

Family history of stuttering

The background questionnaire also included a question about family history of stuttering. Families responded 'yes', 'no' or 'not sure' and were asked to indicate whether family members had continued to stutter into adulthood. Responses were coded as follows: no = 0, yes = 1 (positive family history of stuttering) or not sure = 2.

Analyses

The main objective of the present study was to analyse the influence of child (stuttering severity), family (family history of mental health and stuttering) and contextual (bullying victimisation) factors on RCADS-25-C subscale scores.

All analyses were performed in R version 4.0.3 (R Core Team, 2020). The primary outcome was raw scores on the RCADS-25-C anxiety and depression subscales, which were used in all analyses ($n = 35$). To indicate whether scores reached clinical threshold, mean T-scores, which take into account the child's sex and US school grade, are also reported for this sample. T-scores could be calculated for only 34 participants as sex information was missing for one participant. Scores above 70 are considered to be above clinical threshold, while those between 65 and 70 are considered borderline (Ebesutani et al., 2012).

Predictor variables of interest included age, sex, stuttering severity, IBS score, socio-economic status and family history of adverse mental health and stuttering. To test the strength of the association between the outcome variables and continuous dependent variables and check for multicollinearity amongst predictors ($r > .80$), Pearson's Product Moment Correlation Coefficients were generated using the *corr.test* function in the *psych* Package in R (Revelle, 2021). In order to investigate whether family history variables were associated with symptoms scores, the sample was divided into two groups: positive and negative family history of (i) stuttering and (ii) mental health. Participants who reported 'don't know' or 'prefer not to say' were omitted from the analyses. Welch two sample *t*-tests compared subscale scores in the group of children with a family history of stuttering and the group with no family history.

Multiple regression models were fitted to the data to examine whether these family, contextual, and child variables predicted anxiety or depression subscale scores. The regression models were fitted using the *lm* function in R. The *lm* function automatically generates dummy variables for categorical variables, and family history variables were entered as a dummy variable with two levels (0 = negative family history; 1 = positive family history).

Results

Table 10 shows characteristics of participants included in the sample.

Table 10: Participant characteristics (n = 35)

Participant characteristics (n = 35)				
	<i>Mean</i>	<i>SD</i>	<i>n</i>	<i>% of sample</i>
Age (months)	128	20	35	-
Socio-economic status (IDACI Rank)	17869	8404	30	-
Approximate age of stuttering onset (years)	3.69	1.48	35	-
Sex				
Female	-	-	9	25.7%
Male	-	-	25	71.4%
First Language English	-	-	35	100%
Family history of mental health	-	-	18	51.4%
Yes	-	-	17	48.6%
No	-	-	3	8.6%
Previous access to mental health services	-	-	3	8.6%
Family history of stuttering	-	-	18	51.4%
Ever accessed Speech and Language Therapy for stuttering	-	-	35	100%
Diagnosis of stuttering confirmed	-	-	31	88.6%

Anxiety and depression symptoms

Table 11 presents scores on all child-reported measures for the total sample and by sex. Results of Welch two sample *t*-tests indicated there were no differences in mean scores by sex for anxiety ($t(10.91) = 0.07$, 95% CI [-6.01, 6.43], $p = .94$) or depression symptom scores ($t(18.81) = -1.47$, 95% CI [-4.74, 0.83], $p = .16$). The whole sample was included in all subsequent analyses.

Mean T-scores ($n = 34$) were below clinical threshold (Ebesutani et al., 2012). One participant obtained a T-score above clinical threshold on the anxiety subscale. All T-scores for the depression subscale were within the normative range.

Variables associated with anxiety or depression scores

Child-reported symptom scores were examined in relation to sex, socio-economic status, age, bullying frequency, stuttering severity and family history variables.

Associations between continuous variables

The correlation matrix (Table 12) reports Pearson's product moment correlation coefficients for pairwise comparisons to estimate the strength of association (r) between RCADS-25-C total score, child-reported anxiety, child-reported depression, IBS victim scores, stuttering severity, age and socio-economic status. R-values can be interpreted as effect sizes (Funder & Ozer, 2019), and r -values > 0.20 are likely to be clinically important. Correlation coefficients that remained 'statistically' significant ($p < .05$) after applying the more conservative Holm adjustment for multiple tests are indicated in bold in the table.

Anxiety subscale scores. A moderate, positive correlation was found between anxiety scores and IBS victim scores ($r = 0.52$, 95% CI [0.23, 0.73], $p < .01$, $n = 35$), suggesting that the more bullying a child reported, the higher their anxiety score. This effect remained statistically significant following application of Holm's adjustment. Positive correlations were also found between anxiety scores and self-perceived stuttering severity, ($r = 0.36$, 95%CI[0.03, 0.62], $p = .04$, $n = 35$) as well as anxiety symptoms and age ($r = 0.36$, 95%CI [0.02, 0.62], $p = .04$, $n = 35$).

Depression subscale scores. There was a moderate positive correlation between depression and age ($r = 0.40$, 95%CI [0.07, 0.64], $p = .02$, $n = 35$) indicating that depression symptoms increased as children got older. Weak

correlations were obtained between depression scores and stuttering severity ($r = 0.18$, 95%CI [-0.17, 0.48], $p = .31$, $n = 35$) and IBS victim scores ($r = 0.25$, 95%CI [-0.09, 0.54], $p = .15$, $n = 35$). Here very wide confidence intervals that include zero are observed.

Table 11: Summary statistics for all outcome and predictor variables: RCADS-25-C total and subscale scores, IBS victim scale and stuttering severity rating for the total sample (n=35) and by sex.

	Anxiety subscale		Anxiety T-score		Depression subscale		Depression T-score		IBS victim scale		Stuttering severity	
	M (range)	SD	M (range)	SD	M (range)	SD	M (range)	SD	M (range)	SD	M (range)	SD
Total sample (n = 35)	9.74 (1 – 23)	5.93	44.07 (29.99 – 73.36)	9.39	5.8 (0 – 16)	3.92	43.84 (29.92 – 62.72)	8.86	2.69 (0 – 12)	3.25	6.06 (3 – 10)	1.95
Female (n = 9)	9.89 (1 – 23)	7.82	45.29 (29.99 – 73.36)	14.50	4.44 (0 – 10)	3.13	40.57 (29.92 – 56.51)	8.67	2.78 0 – 12	3.83	4.67 (3 – 7)	1.66
Male (n =25)	9.68 (3 – 23)	5.43	43.63 (34.26 – 62.48)	7.09	6.4 (1 – 16)	4.14	45.02 (32.54 – 62.72)	8.80	2.68 (0 – 12)	3.17	6.56 (3 – 10)	1.87
Undisclosed (n = 1)	10.0	-	-	-	3.0	-	-	-	2.0	-	6.0	-

Note. RCADS-25 subscale score ranges: anxiety (0 – 45) and depression (0 – 30). RCADS-25 T-scores above 70 indicate clinical levels. Data unavailable to calculate T-score for participant who did not disclose sex. IBS Victim subscale score range (0 – 16).

Bullying and stuttering severity. Two thirds of children (65.7%) reported that they had experienced bullying over the past month, although scores indicated frequency of victimisation varied considerably. A moderate, positive correlation was found between bullying score and stuttering severity ($r = 0.38$, 95%CI [0.06, 0.63], $p = .02$, $n = 35$), such that higher self-perceived stuttering severity was associated with more bullying.

Table 12: Correlation Matrix showing Pearson's Correlation Coefficients (r) unadjusted for multiple comparisons. Values in bold indicate that correlation coefficient remained significant after applying Holm's correction for pairwise associations.

	1	2	3	4	5	6	7
1. RCADS-25 Total	-						
2. Anxiety	.93**	-					
3. Depression	.84**	.59**	-				
4. IBS victim scale	.46*	.52*	.25	-			
5. Stuttering Severity	.32	.36*	.18	.38*	-		
6. SES (IDACI)	-.17	-.13	-.18	.14	-.08	-	
7. Age (months)	.41*	.36*	.40*	.29	.20	.11	-

Note: ** $p < .01$, * $p < .05$. Correlation coefficients based on total sample size ($n = 35$) for all variables, except IDACI ($n = 30$) due to missing data. IBS = Illinois Bullying Scale; SES = socio-economic status; IDACI = Income Deprivation Affecting Children Index rank

Relationship between family history variables and RCADS-25 symptom scores

Mean scores by family history group are presented in Table 13. Two sample t -tests were performed to compare subscale scores in children who reported a positive ($n = 18$) or negative ($n = 16$) family history of stuttering.

There was no significant difference in anxiety scores between children who reported a family history of stuttering and those who did not ($t(31.54) = -0.65$, $p = .52$, 95%CI [-5.42, 2.81]). Similarly, groups did not differ in depression scores ($t(29.42) = 0.38$, $p = .71$, 95%CI[-2.30, 3.34]).

Table 13: RCADS-25-C subscale scores for family history variables (categorical variables)

RCADS-25 subscales	Anxiety subscale score		Depression subscale score	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Negative family history of stuttering (n=16)	9.25	5.16	6.19	4.34
Positive family history of stuttering (n=18)	10.56	6.58	5.67	3.63
Negative family history of mental health (n=17)	7.35	4.29	4.0	3.26
Positive family history of mental health (n=18)	12.0	6.48	7.5	3.81

Regression Analyses

Due to the small sample size, it was not possible to include all hypothetical predictors in the regression models. When pre-registering this study, I had stated that independent variables that significantly correlated ($r = 0.40$) with the outcome variable would be included in the regression model. As indicated in the amended pre-registration, four variables (family history of mental health, IBS score, stuttering severity and age) had been identified as priority variables for inclusion, based on theoretical rationale, should the sample size fall below the anticipated 100 participants. Additionally, the continuous variables were significantly associated with at least one of the outcome variables (anxiety or depression subscale score). I therefore included these four variables in the statistical model.

There was a moderate positive correlation between child anxiety and depression subscale scores ($r = 0.59$, 95% CI [0.32, 0.77], $p < .01$, $n = 35$), which would suggest that children who reported high anxiety symptoms also tended to report high depression symptoms. This finding is consistent with

the broader mental health literature, which has indicated anxiety and depression are highly correlated in adolescents and adults (Brady & Kendall, 1992; Lewinsohn et al., 1997). As these variables were only moderately correlated, the outcome variables for the regression analyses were the anxiety and depression subscale scores, rather than the total RCADS-25-C score. Table 14 presents the results of the multiple regression models for anxiety and depression scores.

Anxiety scores

The multiple regression model, which included all four predictors (model 1), was a significant fit to the data ($F(4, 30) = 5.92, p = .001, \text{adj. } R^2 = 0.37$), accounting for approximately 37% of the variance in anxiety scores. Family history of mental health was found to significantly predict anxiety scores ($\beta = 3.87, t = 2.32, p = .03$), suggesting that a positive family history of adverse mental health is associated with a 3.87 point increase in anxiety scores. Observation of the standardised regression coefficients indicates that bullying is also an important factor when considering risk for anxiety in this population ($\beta = 1.82, t = 1.95, p = .06$). Removing age from the model indicated that only IBS scores significantly predicted anxiety scores in this model ($\beta = 2.27, t = 2.43, p = .02$). This model (model 2) was also a significant fit to the data ($F(3,31) = 6.34, p = .002, \text{adj. } R^2 = 0.32$), accounting for 32% of the variance in anxiety scores. Comparison of these two models indicated that the simpler model (model 2) was a better fit to the data ($p=.08$)

Depression scores

The multiple regression model with depression as the outcome variable and family history of mental health and age entered as predictor variables, was a significant fit to the data ($F(2, 32) = 10.44, p < .001, \text{adj. } R^2 = 0.36$), accounting for 36% of the variance in depression symptom scores. Both family history of mental health ($\beta = 3.79, t = 3.54, p < .01$) and age ($\beta = 1.72, t=3.17, p < .01$) significantly predicted depression scores, with a positive family history of adverse mental health and a one month increase in age

each associated with a 3.79 point and 1.72 point increase in subscale scores, respectively.

Table 14: Multiple regression models for anxiety and depression symptom scores (RCADS-25-C). Standardised coefficients for continuous predictors are mean-centred and scaled by 1 standard deviation.

Model	Effect	Standardised coefficients			
		B	SE	<i>t</i> -value	<i>p</i>
Anxiety Models					
Model 1	(Constant)	7.75	1.17	6.61	< .01
	Family history of mental health	3.87	1.67	2.32	.03
	Stuttering severity	0.93	0.88	1.05	.30
	IBS score	1.82	0.93	1.95	.06
	Age (months)	1.56	0.86	1.81	.08
Model 2	(Constant)	8.01	1.21	6.65	< .01
	Family history of mental health	3.37	1.71	1.97	.06
	Stuttering severity	1.10	0.91	1.21	.24
	IBS score	2.27	0.93	2.43	.02
Depression Model					
Model 1	(Constant)	3.85	0.76	5.04	< .01
	Family history of mental health	3.79	1.07	3.54	< .01
	Age (months)	1.72	0.54	3.17	< .01

Discussion

The aim of the present study was to investigate the extent to which contextual, child, and family factors may be associated with self-reported anxiety and depression symptoms in children and adolescents who stutter.

First, I considered whether family history of mental health conditions was associated with increased child-reported symptoms of anxiety or depression. In line with my hypotheses, participants with a family history of mental health difficulties had significantly higher anxiety and depression scores. This is an important finding highlighting the need for future studies comparing anxiety in children who do and do not stutter to account for such familial factors. This would help to tease apart the extent to which the experience of stuttering contributes to anxiety symptoms relative to other putative risk factors for adverse mental health. Another consideration is whether parent-reported anxiety relates to the child's communication challenges or is a pre-existing, longer term problem. For example, it may be that heightened anxiety symptoms may be explained by the genetic risk for anxiety and stuttering per se is not a contributing factor. Alternatively, having a child with communication difficulties may increase parent anxiety, which could have an impact on the child.

Next, I asked whether family history of stuttering acted as a protective factor, attenuating child-reported symptoms of anxiety and depression. Contrary to my predictions, child-reported symptom scores on anxiety and depression subscales did not differ according to family history of stuttering.

Finally, I considered the role of bullying in symptom severity of anxiety and depression for young people who stutter. Anxiety symptom scores were moderately correlated with IBS scores; this association was statistically significant even after adjustment for multiple comparisons. Anxiety symptom scores were also moderately correlated with stuttering severity and age. Although the p -value did not reach conventional levels of statistical significance after adjustment for multiple comparisons, they still represent a medium effect size (Cohen 1988, 1992). The small sample size and heterogeneous profiles in this study likely affected the ability to detect significant effects, and therefore there is need for replication in a larger sample. It was not possible to conduct the proposed mediation analysis as

the sample size was too small, and therefore this remains a question for future research.

In sum, the main finding from this study is that family history of adverse mental health is an important factor when assessing risk for internalising problems in this population. The anxiety model accounted for 37% of the variance in scores. Although, when age was removed from the model, the bullying score was found to be the only significant predictor of anxiety scores. This may suggest an interaction between these variables, and that effects may differ across age. The present study design does not permit examination of these interactions, but there is a need for future studies that employ larger samples and wider age ranges to explore how these contextual, child, and family factors interact at different ages.

The depression model accounted for 36% of the variance in scores and indicated that both family history of mental health and age significantly predicted scores. As evidenced in the literature, prevalence of depression increases with age in the general population, and therefore both of these putative risk factors for depression in the non-stuttering population are also reflected in our sample.

These findings have both theoretical and clinical implications. First, future research comparing anxiety in children who do and do not stutter should account for the influence of family and contextual factors that may moderate the association between anxiety, depression, and stuttering. This would help to enhance our understanding of the relationship between stuttering and internalising problems, and the extent to which the experience of stuttering contributes to risk for elevated anxiety and depression relative to putative risk factors associated with internalising problems. Second, these findings point to the need for clinicians to be alert to both child and broader familial factors associated with adverse mental health when examining risk for poor psychosocial outcomes in children who stutter, to inform ongoing management.

Family history of mental health and stuttering

In the present study, children who had a family history of mental health problems reported significantly higher anxiety and depression symptom scores compared to children who did not disclose a family history of adverse mental health. This is not particularly surprising given that family history of both anxiety and depression are acknowledged risk factors for development of both in children (van Santvoort et al., 2015). Parental anxiety and depression are associated with development of internalising symptoms and disorders in children as a result of both genetic and environmental factors that can have both a direct and indirect impact on children's emotional development (Bayer et al., 2006; Smith, 2004; Thapar et al., 2012). Nonetheless, in the present sample all but one child reported symptoms that did not reach clinical threshold, perhaps indicating that at this age, there is not an association between family history of adverse mental health and clinical levels of internalising symptoms.

There were no significant differences in mean scores of participants who reported a family history of stuttering and those who did not. It was hypothesised that increased experience of the condition would facilitate greater understanding and parental support, which may help to offset any adverse effect on mental health. The current findings do not support the hypothesis that having a parent who stutters acts as a protective factor in the psychosocial impact of stuttering on young people. However, it must be borne in mind that positive family history could include immediate and extended family members, affecting the extent to which children may have had direct experience of stuttering. These data cannot tell us anything about self-perceptions of parents who stutter.

Bullying victimisation

The mean IBS score indicated reasonably low frequency of bullying victimisation in this sample, which seems to contrast with previous studies that found increased risk of bullying in children and adolescents who stutter (e.g. Blood & Blood, 2004; Blood & Blood, 2007; Langevin et al., 1998). This

may relate to the bullying tool used in the present study, which was not designed to tap the unique experiences of children who stutter. Another possibility is that the pandemic disrupted school attendance and therefore perhaps asking children to reflect on bullying experiences over the past month did not capture typical peer experiences as they were not attending school. Nevertheless, two thirds of children reported that they had experienced bullying behaviour over the last month, with varying frequency.

A significant, moderate correlation was observed between self-reported anxiety scores and IBS scores, which is consistent with findings of previous research in the stuttering population (Blood & Blood, 2007; Cook & Howell, 2014). It is well documented that individuals who experience bullying are at heightened risk of adverse mental health, particularly elevated anxiety and depression, in the general population (Moore et al., 2017; Reijntjes, et al., 2010; Scholer et al., 2018). However in the present study, depression symptom scores were not correlated with bullying scores.

It seems reasonable that one may expect there to be an association between risk of bullying and stuttering severity given previous studies showing peer perceptions become more negative as stuttering severity increases (Bloodstein et al., 2021) and the fact that the overt speech characteristics may be more noticeable. On the other hand, Langevin et al. (1998) found that frequency of bullying was not causally related to stuttering severity using a tool designed to examine teasing and bullying experienced by children who stutter specifically. I found a moderate correlation between self-reported stuttering severity and victim subscale scores, although this finding requires replication in larger scale studies.

Stuttering severity

Participants varied considerably in ratings of self-perceived severity. Stuttering severity was correlated with anxiety, but not depression, symptom scores. However, following adjustment for multiple comparisons, this was no longer significant, which is consistent with many published studies that have

similarly reported no significant association between stuttering severity and anxiety in children and adolescents, as discussed in chapter two. For example, previous investigation of anxiety in a larger sample ($n = 96$) of similarly aged children who stutter (9 – 14 years), did not find a significant association between %SS and state ($r = 0.115$) or trait ($r = 0.045$) anxiety (Craig & Hancock, 1996).

Nevertheless, the size of the effect is still clinically important, which leads to the tentative suggestion that, based on the present sample, the more severe a child perceived their stutter to be, the more anxiety symptoms they reported. Further examination of the association between stuttering severity and anxiety, involving a larger sample and different measures of stuttering severity, is needed.

Stuttering frequency is not the only factor associated with severity, although it often is a significant contributor (Manning & DiLollo, 2018). Manning and Beck (2013) argued that measures of the overt features of stuttering, such as frequency, do not account for the broader experience of stuttering, and suggested this may explain the lack of association between stuttering and anxiety reported in studies. In their study, they found that stuttering severity, measured by %SS and the SSI, was not associated with anxiety in adults who stutter ($n = 50$), but scores on the Overall Assessment of the Speaker's Experience of Stuttering (OASES) were associated with anxiety (Manning & Beck, 2013). The OASES is an alternative measure of stuttering severity in that it assesses the broader impact of stuttering. Interestingly, Manning and Beck (2013) found no significant association between depression and any measure of stuttering severity (%SS, SSI, OASES), which is consistent with our findings.

In the present study, I employed a self-report rating scale of stuttering severity guided by the rationale that it might offer greater insight into the child's perceptions of their own speech. While this is not an objective measure of frequency, the extent to which a child perceives their speech to be dysfluent may be associated with self-reported experience of anxiety

symptoms. Nonetheless, inclusion of the OASES would have been a valuable addition to the present study and would have permitted comparison of the association between anxiety and stuttering severity measured in two different ways. Limitations associated with data collection during the pandemic meant that I opted for a shorter, simpler measure to minimise missing participant data.

Results of the regression analyses indicated that stuttering severity did not significantly predict anxiety subscale scores. These findings contrast with a larger study ($n = 102$) involving adolescents seeking treatment for stuttering, which found that higher self-rated stuttering severity predicted higher anxiety and internalising problems (Iverach et al., 2017b). One possible reason for divergent findings may relate to sample selection. The fact that participants were seeking treatment may suggest that their stutter had a greater functional impact, influencing perceived severity and self-reported anxiety. It should also be borne in mind that the study by Iverach and colleagues (2017b) comprised an older sample (11 – 17 years). The length of time one has lived with a stutter may influence the association between stuttering severity and internalising symptoms. Therefore, future studies involving community samples may provide further insight into factors that predict internalising symptoms.

Stuttering severity, bullying and internalising symptoms

This study also aimed to address the question of whether bullying mediates any relationship between stuttering severity and anxiety and/or depression symptoms. This question could not be investigated as a sufficient sample size was not achieved, and the study design lacked the necessary temporal elements to reliably estimate a mediation effect. Instead, larger, and preferably longitudinal, studies are needed to adequately address this research question. The present results do suggest that bullying victimisation likely contributes to heightened anxiety symptoms, although there is a need for future research to consider how these contextual and child factors interact over time. Previous research has indicated that bullying reported in this population is often related to an individuals' stutter (Erickson & Block, 2013;

Langevin et al., 1998). Furthermore, population studies of non-stuttering cohorts have shown bullying victimisation to be associated with anxiety and depression, therefore these findings highlight the need for families, teachers and clinicians to be aware of the elevated risk for adverse mental health in children who stutter who are subjected to bullying.

Clinical implications

The present findings indicate there is a need for clinicians to take into account family and contextual factors in assessment and management of children who stutter, as well as child factors. In particular, clinicians need to be aware of the elevated risks for internalising problems associated with family history of adverse mental health, and possibly bullying victimisation, when planning ongoing management. Mental health screening may indicate need for multidisciplinary involvement, such as referral to Psychology Services, or indicate a need to incorporate psychological therapies to help offset development of anxiety in children identified as at risk. Consideration of risk for internalising problems may be particularly pertinent among adolescents given that prevalence of anxiety and depression increases with age in non-stuttering adolescents, and age was a significant predictor of depression in the present study. However, longitudinal community studies are needed to improve our understanding of how these child, family, and contextual factors interact with each other across development.

While appropriate clinical management may help to offset risk for internalising problems associated with familial factors, initiatives which address contextual factors found to detrimentally affect emotional well-being are also worthy of consideration. In particular, these findings underscore the role of schools, health services and charities in alleviating risk of bullying in young people who stutter. This could involve, for example, anti-bullying initiatives, awareness and understanding about stuttering in society and efforts to tackle stigma. Programs focused on stuttering education and bullying awareness have shown potential for facilitating positive changes in

peer attitudes towards children who stutter and bullying (e.g. Langevin et al., 2012).

Limitations

Most participants in this study completed the questionnaire during the global coronavirus pandemic (from March 2020), which must be taken into consideration when interpreting the findings. Research examining the impact of the pandemic on child and adolescent mental health has reported a mixture of negative and positive experiences, which are likely associated with a number of risk and protective factors (Panchal et al., 2021). For example, a recent systematic review has shown female sex and adolescence (13 – 15 years) were significantly associated with elevated anxiety and depression during lockdown (Panchal et al., 2021). Social stresses and adverse events are known risk factors for development of anxiety and depression in young people. It should be recognised that some of these environmental risk factors will have been experienced by an unprecedented number of families during the pandemic, for example bereavement, family discord, financial pressures and broader social stressors. In spite of the impact of the pandemic on everyday life, this sample did not show clinical levels of anxiety or depression. Nevertheless, symptom scores reported in the present analyses are likely confounded by the experiences of the global coronavirus pandemic, and as such are not comparable with population norms.

I had intended to recruit 100 participants to this study, which should have permitted robust analysis of the original seven predictors. However, recruitment was challenging given the pandemic context and the final sample size was comparatively smaller, resulting in reduced statistical power. It is important to note the wide confidence intervals associated with many of the correlation coefficients in the reported analyses, suggesting a reasonable amount of variation within the group. The small sample size may have resulted in overestimation of the effect size (Funder & Ozer, 2019), and therefore there is need for replication in larger studies. The present sample

may also be affected by selection bias relating to participant treatment status and accessibility of an online study. First, I recruited through word of mouth and digital advertisement of the study, predominantly through the charity's social media channels. All of the children had previously accessed speech and language therapy and therefore may have been more aware of the psychosocial impact of stuttering on children's mental health and well-being, and this may also have been the focus of therapy. Second, an online questionnaire requires access to a computer and stable internet connection preferably at home. The study therefore automatically excluded those participants who could not access an online study. Third, families who were experiencing unprecedented stresses due to the novel pressures of the pandemic may have been less likely to participate, skewing the sample.

Finally, recent studies have found that children and adolescents who stutter who have co-occurring neurodevelopmental conditions present with elevated anxiety and emotional difficulties compared to those who stutter without additional difficulties (Blood et al., 2007; Briley et al., 2019; Smith et al., 2017). A limitation of the present study is that presence of co-existing diagnoses amongst participants is unknown. This will be an important variable to include in future studies that employ community samples of children and adolescents who stutter as this can inform our understanding of factors that influence anxiety and possibly depression in this population.

Conclusion

The present study aimed to evaluate the association between anxiety and depression symptoms and several child, familial, and contextual factors in a cohort of children who stutter. Family history of adverse mental health was a significant predictor of anxiety and depression symptoms. Age was also found to significantly predict depression scores. In spite of the limitations, particularly the confound of the COVID-19 pandemic on population mental health, these findings contribute further evidence that there is a need for clinicians to consider a number of family, child, and contextual factors in risk for anxiety in school-age children who stutter. This finding offers an

opportunity for schools and clinicians to champion anti-bullying initiatives that could help prevent the negative psychosocial impact of bullying victimisation in stuttering. Future research is needed to explore further factors associated with elevated anxiety and depression in children and adolescents who stutter, and how these factors may interact across development, using longitudinal population studies.

Chapter 5

Internalising symptom trajectories in children who do and do not stutter

Abstract

Purpose. Questions remain about the psychosocial development of children and young people who stutter and factors that may serve to increase risk in this population. This study aimed to investigate within-person (intra-individual) change in internalising symptoms over time in children reported to stutter, children with other speech and language difficulties, and those with typical speech and language skills. The cumulative risks that may serve to elevate anxiety and depression symptoms in adolescents who stutter were also examined.

Method. Data were obtained from the Millennium Cohort Study, a large UK birth cohort ($n = 17,256$). Latent growth curve models were fitted to parent-reported scores on the Strengths and Difficulties Questionnaire (SDQ) across six time points (3 – 17 years). The associations between stuttering and self-harm and anxiety or depression diagnosis at age 17 were investigated using multiple logistic regression models. Multiple risk factor models examined the effect of predictors on clinical-level symptoms in the stuttering sample ($n = 565$).

Results. After controlling for speech and language problems, cohort members who stutter had higher internalising symptom scores at age three ($b = 0.33, p = .047$) compared to non-stuttering peers, but symptom trajectories did not differ relative to non-stuttering peers over time. Cohort members who stutter and had speech and language problems had the highest internalising scores, followed by those with speech and language problems only. After adjusting for child sex, ethnicity, verbal ability, co-occurring speech and language problems, autism or ADHD diagnosis, and SES, stuttering no longer had an effect on internalising scores. However, cohort members with speech and language problems had higher internalising symptoms at age 3 and differed in the rate of change over time compared to

those without speech and language problems. There was also evidence of an additive effect of stuttering and speech and language problems, in so far as these cohort members had the highest symptom scores at age three and over the course of development. Stuttering was not associated with increased odds of self-harm or clinical anxiety or depression. Female sex and maternal mental health were associated with increased odds of scoring above threshold on mental health measures in the stuttering sample.

Conclusion. These findings suggest there are no differences in the internalising symptom trajectories of children who do and do not stutter, once child- and family-factors are taken into account. Speech and language problems may be associated with higher internalising symptoms, and clinicians should be alert to the possible additive effect of co-occurring difficulties and stuttering. Clinicians need to be aware of elevated risk for internalising problems associated with co-occurring communication difficulties and female sex among children who stutter.

Introduction

Internalising disorders, which include anxiety and depression, are a group of conditions characterised by disordered emotion or mood (Achenbach, 1966; Kovacs & Devlin, 1998). It is widely recognised that poor mental health in young people poses a substantial public health challenge (Liu, Chen & Lewis, 2011; Patel et al., 2007), with implications for broader health, education and social functioning (Costello, Egger & Angold, 2005; Liu, Chen & Lewis, 2011; Patel et al., 2007; Pine et al., 1998). Furthermore, experience of anxiety and depression during adolescence is associated with increased risk for recurrence in early adulthood (Copeland et al., 2021; Pine et al., 1998). Internalising problems have reportedly been increasing in adolescents, particularly amongst girls, in high income countries (Bor et al., 2014; Collishaw, 2015). In the UK, increasing rates of anxiety and depression have been observed in children and adolescents in recent decades (Cybulski et al., 2021; Patalay & Gage, 2019).

Risk factors associated with internalising problems in adolescents

There are a number of factors associated with elevated risk for developing anxiety or depression in the general population. These include parental mental health problems, inhibited temperament, low socio-economic status, peer relationship difficulties, bullying victimisation, negative family relationships, adverse childhood experiences, and female sex (Fryers et al., 2002; Green & Benzevel, 2013; Narmandakh et al., 2020; Pine & Klein, 2015; Rapee, 2015; Thapar et al., 2012). Some studies indicate that mental health outcomes, and internalising symptom trajectories, differ between ethnic groups in the UK (Bains & Gutman, 2021; Goodman, Patel & Leon, 2008). In the latest pre-pandemic Mental Health in Children and Young People in England (2017) survey, emotional disorders were more common in girls, children of parents with mental health issues, young people from lower socioeconomic groups, and children of White British and mixed ethnic backgrounds (Sadler et al., 2018). A longitudinal birth cohort study following

over 54,000 young people aged 10 – 21 years concluded that anxiety and mood disorders share several common risk factors, while parental history of either disorder and female sex were identified as the strongest predictors of both (Hyland et al., 2016). Family history of depression and psychosocial stress are considered the strongest risk factors for depression in adolescents (Thapar et al., 2012).

Childhood speech, language and communication disorders are associated with increased risk of adverse mental health, including emotional difficulties, and heightened vulnerability to problems with peers, friendships, and bullying (Wren et al., 2023; Durkin & Conti-Ramsden, 2010; Keating, Turrell & Ozanne, 2001; Prizant et al., 1990). Compared to the general population, risk for elevated anxiety and depression has also been associated with a number of neurodevelopmental conditions, including attention deficit hyperactivity disorder (ADHD), autism spectrum conditions, developmental dyslexia, and language disorder (Gnanavel et al., 2019; Hurtig et al., 2009; Kim et al., 2000; Livingston, Siegel & Ribary, 2018; Skokauskas & Gallagher, 2012; White et al., 2009; Yew & O’Kearney, 2013).

Stuttering and internalising symptoms

Developmental stuttering is one such neurodevelopmental condition that has long been associated with internalising problems, particularly anxiety.

Stuttering is a speech condition characterised by overt behaviours that result in disruptions to the rhythm and timing of speech, including repetitions, prolongations and blocks in airflow, and concomitant behaviours, such as involuntary movements and word avoidance or substitution. These more observable characteristics may be accompanied by affective and cognitive reactions, for example negative feelings, emotions and attitudes (Bloodstein, Bernstein Ratner & Brundage, 2021). Children and adolescents who stutter exhibit more negative communication attitudes, heightened communication apprehension and poorer self-perceived communication competence relative to peers who do not stutter (Blood et al., 2001; Guttormsen, Kefalianos & Næss, 2015).

Many individuals who stutter report facing public stigma, including negative reactions and social devaluation, which has been found to negatively correlate with mental health outcomes in adults (Boyle, 2018). On the other hand, some studies have shown that adolescents who stutter do not necessarily perceive stuttering to be a stigmatising condition nor that it affects likelihood of peers wishing to be friends (Blood et al., 2003b; Erickson & Block, 2013). Nevertheless, studies have indicated that children and adolescents who stutter are at heightened risk of bullying and experience negative peer reactions (Blood & Blood, 2004; Blood & Blood, 2007; Davis, Howell & Cooke, 2002; Erickson & Block, 2013; Langevin et al., 1998). Among young people who stutter, childhood bullying has been associated with self-reported anxiety, low self-esteem, reduced life satisfaction and poor self-perceived communication competence (Blood & Blood, 2007; Blood & Blood, 2004; Blood et al., 2011; Blood & Blood, 2016). Consequently, the negative experiences and communication challenges experienced by individuals who stutter may increase risk for anxiety in adolescents and adults who stutter (Iverach & Rapee, 2014; Smith et al., 2014).

In chapter two, conclusions from the meta-analysis suggested that stuttering may be associated with increased risk for anxiety in some school-aged children, although findings across included studies were variable, which may be partly attributable to several moderating factors. The relationship between stuttering and depression is less clear, although studies to date have failed to find an association between depression and stuttering in children and adolescents (Bernard, Hofslundsengen & Norbury, 2022; Livingstone-Pountney & Mitrevski, 2019). Determining whether children who stutter are at elevated risk of internalising symptoms has implications for clinical management in terms of awareness, early identification, and prevention of adverse social, emotional and educational consequences.

In their narrative review, Smith et al. (2014) examined a number of putative cognitive, environmental and familial risk factors associated with anxiety in relation to stuttering. Smith et al. (2014) concluded that there was limited

evidence that children who stutter have a genetic predisposition to anxiety or exhibit temperament traits often considered precursors. Instead, it has been hypothesised that environmental factors, such as increased risk of bullying, negative peer reactions, and stigma contribute to adverse mental health in the stuttering population (Boyle, 2018; Smith et al., 2014).

Nevertheless, there appears to be limited consideration of these putative risk factors for mental health in studies looking at anxiety in the stuttering population, even though such factors may serve to increase risk (Bernard, Hofslundsengen & Norbury, 2022; Smith et al., 2017). The fact that studies to date have highlighted the heterogeneity amongst children who stutter in the extent to which they report elevated internalising symptoms perhaps indicates that a number of factors, both intrinsic to and independent of stuttering, may play a role in development of anxiety and/or depression in this population. For instance, as cited in chapter two, few studies of anxiety in stuttering cohorts controlled for co-occurring communication and/or learning difficulties; those that did found that children who stutter who have co-occurring communication difficulties report higher anxiety symptoms than children who stutter without additional needs (Blood et al., 2007; Briley, O'Brien & Ellis, 2019). Consequently, there is a need for further investigation of factors that may serve to increase risk for poor mental health, which may contribute to our understanding of the individual differences in child outcomes.

Development of internalising symptoms over time

Understanding the relationship between stuttering and mental health has been hampered by reliance on small samples recruited from clinical services (Bernard, Hofslundsengen & Norbury, 2022; Craig et al., 2003; Menzies et al., 1999; Smith et al., 2014). Reliance on clinically ascertained samples may overestimate levels of anxiety in the stuttering population as these children may differ from children who do not seek intervention (Craig et al., 2003). In addition, intervention may, directly or indirectly, affect feelings and attitudes

around stuttering, again influencing anxiety levels in those who have received treatment (Craig, 1994).

Consequently, studies employing community samples are likely to yield a more representative estimate of risk for elevated anxiety in children and young people who stutter. Two such community cohort studies in the UK and Australia failed to find evidence that school-aged children who stutter present with elevated anxiety symptoms relative to non-stuttering peers (Andrews & Harris, 1964; Smith et al., 2017).

However, few community cohort studies include longitudinal designs documenting symptom changes over time. Studies that have drawn on longitudinal data have tended to make cross-sectional comparisons of anxiety symptoms in children who do and do not stutter rather than plotting symptoms over time in the same individuals. McAllister (2016) analysed data from three sweeps of the Millennium Cohort Study (MCS), to examine the social, emotional and behavioural development of children who stutter compared to typically developing peers. After controlling for confounding factors, significantly higher scores were reported for the stuttering group on all subscales of the Strengths and Difficulties Questionnaire (SDQ) at ages five and 11 years. As McAllister acknowledges, the children in each sweep were not necessarily the same children and therefore this study cannot illustrate within-person change over time.

The Early Language in Victoria Study (ELVS) is a longitudinal, population-based cohort study that has collected multi-informant data about language development from infancy (8 months) through to adolescence in Australia (Reilly et al., 2018). The prospective longitudinal design used in the ELVS stuttering study offers insight into socio-emotional outcomes of children who stutter at different ages. For example, Kefalianos et al., (2014) found that children who stutter ($n = 183$) aged 2 – 4 years did not exhibit temperament traits identified as precursors to anxiety relative to the non-stuttering comparison group. At age seven, parent-reported psychosocial differences were not observed between the stuttering and non-stuttering groups, nor

between children classified as persistent ($n = 34$) or recovered ($n = 67$) stuttering status (Kefalianos et al., 2017). At age 11 years, Smith et al. (2017) compared anxiety symptoms on multiple self- and parent-report measures using the ELVS dataset. For this analysis, the stuttering cohort ($n = 141$) was divided into persistent ($n = 20$) and recovered ($n = 121$), and included a non-stuttering comparison group ($n = 702$). Children with a diagnosis of autism or learning difficulties were included in this sample. After controlling for co-occurring diagnosis of learning difficulties or autism, adjusted analyses indicated no evidence that children who persisted in stuttering were more anxious than children who recovered or who did not stutter, though the numbers in the persistent stuttering group were small.

Both the ELVS and MCS studies have provided valuable cross-sectional insight into the psychosocial outcomes of school-aged children who stutter based on community cohorts, but these studies have not considered changes in symptom profiles that may occur during adolescence. Questions therefore remain concerning the psychosocial *development* of children and young people who stutter during the transition to adolescence, when onset of internalising symptoms peak (Maughan & Collishaw, 2015). This research aims to build on existing work by using longitudinal data analysis to investigate within-person (intra-individual) change over time in a group of children reported to stutter by their parents.

As discussed, children who experience language and communication difficulties are at increased risk for internalising symptoms in adolescence (Yew & O'Kearney, 2013). This gives rise to two further questions. First, is stuttering per se associated with elevated emotional symptoms or does presence of developmental comorbidities partly explain the heterogeneity in symptoms reported across studies? It seems reasonable to assume that children with co-existing difficulties are likely to face greater challenges, which may increase their risk for adverse mental health outcomes. Nevertheless, consideration of developmental comorbidities in any association between stuttering and internalising problems requires further investigation. Second, given the documented risk of adverse mental health in

children with other communication problems, does stuttering in the absence of other communication problems pose a unique risk for internalising symptoms? The present study will analyse data obtained from the UK Millennium Cohort Study (MCS) to evaluate risk for internalising symptoms among children and adolescents who do and do not stutter, accounting for potential co-occurring speech, language, and communication difficulties.

Present study

In the present study, internalising symptom trajectories are plotted for the same group of individuals (age 3 – 17 years). The goal is to improve our understanding of the approximate age at which children who stutter may be at elevated risk of emotional problems. I will also examine between-person (inter-individual) change by examining the symptom trajectories of children with and without other parent-reported speech and language concerns in an effort to ascertain whether stuttering poses a unique risk for internalising problems relative to other communication problems. Finally, using the wealth of family, parent- and cohort member-level data in the MCS, I investigate cumulative risk for internalising problems in this population. This will enhance our understanding of the heterogeneity in symptoms, with implications for the clinical management of children who stutter.

The current study was pre-registered with the Open Science Framework in September 2022 (<https://osf.io/wfk5h>). Through the analysis of secondary data from the MCS, I aim to address the following research questions:

- What is the trajectory of internalising symptoms in children and adolescents (3 – 17 years) who stutter in a UK birth cohort?
- Do the internalising symptom trajectories of children who stutter differ significantly from the symptom trajectories of children with parent-reported speech and language problems (other than stuttering) in a UK birth cohort?
- To what extent does presence of multiple risk factors increase risk of elevated internalising problems in adolescents who stutter?

Method

Data

The present study involved analysis of secondary data obtained from the longitudinal Millennium Cohort Study (MCS), which is accessible via the UK Data Archive. The MCS follows 19,244 children born between September 2000 and January 2002 in the UK (Connelly & Platt, 2014). To date, data have been collected at seven sweeps, when cohort members were aged 9 months, 3, 5, 7, 11, 14 and 17 years. At sweep two, the baseline sample ($n = 18,552$) was supplemented with an additional 692 eligible families who had not been included in the first sweep. Further information about the MCS can be found on the Centre for Longitudinal Studies website (<https://cls.ucl.ac.uk/cls-studies/millennium-cohort-study/>). Ethical approval for the present study was granted by University College London Ethics Committee (ID: 15535/02).

Participants

Children eligible for inclusion in the MCS were identified using Child Benefit Records; a universal monetary benefit available to families living in the UK. The MCS uses a disproportionately stratified sample design, whereby typically under-represented groups, including ethnic minority groups, disadvantaged families, and children from the devolved nations (Wales, Scotland, and Northern Ireland), were over-sampled to make the MCS sample representative of the population (Connelly & Platt, 2014). The MCS sample comprises 253 sets of twins and 11 sets of triplets (see Connelly & Platt, 2014). Multiple cohort members from the same family were not included in the present study as the shared experience of growing up in the same family is likely to influence mental health outcomes. As stuttering has high heritability and therefore it is possible that twin children will both stutter, the firstborn cohort member in these families was selected for inclusion to minimise selection bias. Triplet data were not obtained as I did not have appropriate permissions to access these data.

Data for sweeps one to seven have been used in the present analyses. Only cohort members who provided data for the speech, language and communication difficulties question in at least one sweep were retained in the sample, resulting in a total sample size of $n = 17,256$. Although power analyses were not conducted during study design, it was anticipated that a population study comprising such a large sample would result in adequate power to detect meaningful differences between groups. Nevertheless, confidence intervals have been reported in the results section in preference to conducting post-hoc power analyses (Heckman, Davis & Crowson, 2022).

Measures

Predictor Variable: Grouping variables

Two binary grouping variables were the independent variables in analyses: a) does the child stutter (yes/no) and b) does the child have any speech or language difficulties (yes/no). These binary variables were based on responses to the speech, language and communication difficulties question at ages 3, 5, 11 and 14 years (see Appendix F). This question was not asked at age 9-months, seven or 17 years.

At MCS2 and MCS3, the main respondent was asked '*do you have any concerns about the cohort member's speech and language?*', to which they could select the relevant response from a list. No concerns, stuttering, 'language developing slowly', 'doesn't understand others' and 'pronounces words poorly' informed the grouping variables as these options were consistent across the two sweeps. Inclusion of other speech, language or communication concerns listed, such as hearing problems or lisps, introduces variability in the nature of the difficulties represented in the group, potentially confounding analyses.

At MCS5 and MCS6, the main respondent was asked '*does the cohort member have any problems with any of the following?*' and '*has a doctor or health professional ever told you that the cohort member has a problem with any of the following?*', respectively. The options available to respondents in

both sweeps included 'a stutter or stutter', 'another problem with talking', 'a problem with understanding what other people say'.

(i) *Stuttering variable.*

Cohort members were assigned to the stuttering group if the main respondent stated the child had a stutter in at least one sweep. I was interested in the extent to which previous or current experience of stuttering may be associated with internalising symptoms. Adolescence is a time of significant transitions – physically, emotionally, and socially – and therefore the current study examined the longer-term mental health outcomes of individuals reported to stutter at any point in development.

(ii) *Speech and language problems variable.*

The speech and language variable was the second binary predictor variable (1 = speech & language problems; 0 = no speech & language problems). Presence of speech and language problems depended on an affirmative response in at least one sweep.

Due to issues with attrition (i.e. cohort member may not have participated in the subsequent sweep) and reliance on parent-reported difficulties as opposed to clinical assessment, resolution of stuttering or speech and language problems could not be determined from the data available in the MCS. Although stuttering and speech/language difficulties do not necessarily persist to adulthood, experience of either during development may continue to affect children even in instances where the communication issue has resolved. Therefore, the current coding system permitted examination of within- and between-individual differences in those who had ever reported stuttering and/or speech and language problems, compared with those who had not reported either.

Outcome variables: mental health measures

For each sweep, data were obtained through interviews with parents/main caregivers of the cohort member, and in MCS6 and MCS7 data were also

collected from cohort members themselves. Data were collected through main and partner respondent interviews, or proxy partner interviews in instances where the partner was temporarily unavailable. Eligibility to be the main and partner respondents depended on the relationship to the cohort member and between members of the household. Usually, the mother was identified as the main respondent and the father for the partner respondent. In the event parents did not reside in the household, the main carer was selected for interview.

(i) *Strengths & Difficulties Questionnaire (SDQ)*

The Strengths & Difficulties Questionnaire (SDQ; Goodman et al., 2001) is a behavioural screening questionnaire comprising five subscales. When using the SDQ with community samples, it is recommended that researchers consider using the broader internalising and externalising subscales (Goodman et al., 2010).

Parent-reported SDQ scores were available at six time points (MCS2 – MCS7), when cohort members were aged 3, 5, 7, 11, 14 and 17 years. In the MCS, the main respondent is not necessarily the same person across sweeps, which may affect the stability of symptom reporting, making it difficult to reliably plot internalising symptom trajectories over time.

Therefore, SDQ data were filtered by the individual who most frequently responded at each sweep, to ensure that all data on this outcome variable were provided by the same respondent. Self-reported SDQ scores were also available at MCS7 and contributed to the regression analyses.

SDQ internalising scores at each sweep were entered as continuous variables in the latent growth curve models. Scores on the emotion and peer problem subscales were summed to generate the internalising symptom score (range 0 – 20). For logistic regression analyses, SDQ Emotion subscale scores were dichotomised to indicate whether scores fell above or below clinical thresholds. In the SDQ manual, parent-reported emotion subscale scores can be categorised into normal (0 – 3), borderline (4) and abnormal (5 – 10). For the purposes of the present analyses, the borderline

scores were collapsed into the normal range, creating a 'below (0 – 4) and 'above' (5 – 10) threshold score. Self-reported emotion subscale scores are categorised as normal (0 – 5), borderline (6) and abnormal (7 – 10). In the present analyses, self-reported scores were dichotomised into 'below' (0 – 6) and 'above' (7 – 10) threshold.

(ii) *Short Moods & Feelings Questionnaire (SMFQ)*

The Short Moods & Feelings Questionnaire (SMFQ; Angold, Costello & Messer, 1995) is a self-report measure of depressive symptoms in children and young people aged 6 to 17 years. It measures cognitive and affective symptoms over the last two weeks (range 0 – 26). Scores of 12 or greater suggest possible presence of depression. Cohort members completed this scale at MCS6, when they were aged 14 years. For the present analyses, SMFQ scores were categorised as 'above' (≥ 12) or 'below' (≤ 11) threshold (categorical outcome variables).

(iii) *Kessler Psychological Distress Scale (K6)*

The Kessler-6 (Kessler et al., 2003) is a 6-item self-report measure used to evaluate psychological distress and is intended to indicate risk for serious mental illness (range 0 – 24). Scores of 13 or higher indicate severe mental distress. Cohort members' scores were dichotomised as 'above' (≥ 13) and 'below' (≤ 12) threshold. Cohort members completed this scale at MCS7, aged 17 years.

(iv) *Cohort member report of self-harm (binary variable)*

At MCS6 cohort members were asked: '*In the past year, have you hurt yourself on purpose in any way?*'. Responses were coded 'yes' (reported self-harm) and 'no' (did not report self-harm). At MCS7, cohort members responded to the question: '*During the past year, have you hurt yourself on purpose in any of the following ways?*' An affirmative response to at least one of the examples of self-harm listed resulted in the cohort member being assigned to the self-harm group at MCS7.

(v) *Cohort member report of suicide attempt (binary variable)*

At MCS7, cohort members were asked, '*Have you ever hurt yourself on purpose in an attempt to end your life?*'. Responses were coded 'yes' or 'no'.

(vi) *Diagnosis of anxiety or depression*

At MCS7, cohort members were asked, '*Has a doctor ever told you that you suffer from depression or serious anxiety?*' This was a hierarchical question, in which cohort members who affirmed they did have a diagnosis, were asked three more questions:

'At what age were you first diagnosed with depression or serious anxiety?'

'Are you currently being treated for depression or serious anxiety?'

'Have you ever received treatment for depression or serious anxiety?'

Responses were coded 'yes' or 'no' to this initial question.

Covariate variables

Demographic variables

Demographic data (cohort member sex and ethnic group) and family variables (socio-economic status) were obtained from MCS1 and MCS2. Three measures of socio-economic status (SES) were included in this study: Organisation for Economic Cooperation and Development (OECD) below 60% median poverty indicator (above = 0, below = 1); weighted OECD income quintiles (1 = lowest, 5 = highest); maternal National Vocational Qualification (NVQ)-equivalent with six levels (1 = NVQ Level 1 through to 5 = NVQ Level 5, 6 = overseas equivalent). The poverty indicator refers to disposable household income that falls below 60% of the median national income. NVQ level one is equivalent to GCSE (grade D – E) qualifications, and NVQ level five is equivalent to an undergraduate qualification. Data for socio-economic status were obtained at the first two sweeps as the proportion of missing data was lower and onset of stuttering typically occurs around this point.

Cohort member characteristics

Co-occurring autism or ADHD. The main respondent was asked whether the cohort member had been diagnosed with autism or attention deficit disorder (ADHD) at four sweeps (MCS 3 – 6), when cohort members were aged 5, 7, 11 and 14. As autism and ADHD are lifelong conditions with varying age of diagnosis (Mandy et al., 2022), confirmation of diagnosis at any of these four sweeps was used to generate two binary variables (0 = no diagnosis, 1 = diagnosis); one for Autism and one for ADHD.

Bullying. At MCS4 and MCS5, cohort members were asked whether they had experienced bullying. In MCS6, cohort members were asked to indicate if they had experienced bullying or cyberbullying. This information was coded as a binary variable (0 = not bullied, 1 = ever bullied) depending on affirmative response in at least one sweep.

Cognitive ability. Measures of cognitive ability at age 3 years included T-scores on the British Ability Scales II (BAS) Naming Vocabulary Test (verbal ability) and standard scores on the Bracken School Readiness Scale (non-verbal ability), both of which adjust for cohort members' age. These were to be included as covariates in the latent growth curve models to account for broader cognitive development. In the MCS, cognitive measures change across sweeps, making it difficult to compare performance over time. Research has suggested that language trajectories remain relatively stable from around age five years (Norbury et al., 2017), however age three is the first point at which data for our predictor variable were collected.

Parent mental health. Parent (mother and father) mental health was measured using two variables: (i) scores on the Kessler-6 measure of psychological distress for sweep MCS2 – MCS7¹⁰, and (ii) report of a serious anxiety or depression diagnosis at each sweep. For the anxiety or depression diagnosis, data were amalgamated across sweeps MCS1 to MCS7 to generate a binary variable (0 = no anxiety or depression diagnosis; 1 = anxiety or depression diagnosis). Participants were assigned a diagnosis of anxiety or depression if they responded 'yes' at any sweep. Kessler-6 scores were entered as continuous variables in subsequent analyses.

Analysis

Missing data strategy

Multiple imputation by chained equation modelling was implemented to account for the missing data in the final sample, using the *mice* package in R (van Buuren & Groothuis-Oudshoorn, 2011). Data were imputed twenty times, creating five imputed datasets.

Two approaches were employed to deal with missing data on mental health measures. In instances where there was a relatively small amount of missing item-level data (<50% of scale items), the mean item score on that measure for that cohort member was imputed prior to calculating the total score. When data were missing because a cohort member did not complete the measure at all or completed fewer than half of items, the total scores were imputed using the *mice* package in R (van Buuren & Groothuis-Oudshoorn, 2011).

For the present sample, there were no missing data for the predictor variables (stuttering and speech/language problems variables). For information on the proportion of missing data by variable, refer to Appendix F. Design and strata weights were applied to analyses to account for the stratified cluster design and over-sampling. However, the overall weights, which account for sample attrition, were not used with imputed data. All analyses were performed in R version 4.2.1 (R Studio, 2022). Analyses one to three involve the whole sample ($n = 17,256$), and analysis four involves only the stuttering sample ($n = 565$).

Analysis 1: symptom trajectories

Weighted descriptive statistics for continuous variables were estimated using the *mitools* package (Lumley, 2022), which applies Rubin's Rule when pooling estimates from the five imputed datasets to generate a final estimate. Summary statistics for categorical variables reflect the most frequently occurring category assigned across the five imputed datasets.

To examine internalising symptom trajectories over time, latent growth curve models were fitted to parent-reported SDQ internalising scores across six time points (MCS2 – MCS7). The *lavaan* (version 0.6-12; Rosseel, 2012) and *semTools* (Jorgensen et al., 2022) packages were used to fit four latent growth curve models to the multiply imputed datasets. Model 1 was the baseline model, which modelled the mean intercept and slope for the whole sample. The grouping variables (stutter – yes/no; speech & language problems – yes/no) were then included in the conditional model to permit estimation of group differences in symptom trajectories (model 2). Quadratic terms were added into this model to examine non-linear trajectories (model 3). Larger values of the quadratic slope suggest the rate of change is changing more rapidly (Grimm, Ram & Hamagami, 2011). To assess the robustness of this model to potential confounding variables, I report a fourth model adjusted for variables that may be associated with mental health and well-being (sex, ethnic group, SES, parent Kessler-6 score, age three cognitive ability, co-occurring autism or ADHD). Indices to assess model fit include the Tucker-Lewis Index (TLI; $>.90$ indicates good fit), Comparative Fit Index (CFI; greater than $>.90$ acceptable and $>.95$ good fit), and Root Mean Square Error of Approximation (RMSEA; $<.08$ and $p >.05$ good fit) and Standardised Root Mean Residual (SRMR; $<.05$ good fit).

Analysis 2: Risk for self-harm

The association between stuttering and self-harm at age 17 were analysed using logistic regression models. Self-harm was entered as a binary outcome variable (0 = no self-harm, 1 = self-harm). Model 1 (unadjusted) estimated the effect of stuttering on likelihood of reporting self-harm. Model 2 assessed whether stuttering has an effect on risk for self-harm once speech and language problems, sex, SES, ethnic group and parent mental health had been accounted for.

Analysis 3: Risk for anxiety or depression diagnosis

Logistic regression models were fitted to estimate whether stuttering is associated with greater risk for anxiety or depression diagnosis at age 17.

Model 1 estimated the association between stuttering and anxiety or depression diagnosis (binary variable: 0 = no diagnosis; 1 = diagnosis), while Model 2 adjusted for covariates, including speech and language problems, sex, SES, ethnic group and parent mental health.

Analysis 4: multiple risk models

The final set of analyses focused on the association between putative risk factors and internalising problems in adolescence among the stuttering group only ($n = 565$). This was to test the final hypothesis regarding a multiple risk factor model.

A number of statistical models can be utilised to examine the effect of multiple risk factors on child outcomes. The cumulative risk (CR) model is one such approach frequently used in developmental psychology, which research has found to be a robust predictor of children's mental health problems (see Evans, Li & Whipple, 2013; Etekal et al., 2019). The CR model involves aggregating multiple risk factors to form a composite variable or cumulative risk index. In a CR model, risk exposures are coded dichotomously to reflect presence or absence of a risk factor (e.g. 1 = risk, 0 = no risk), and these dichotomous risk values can be summed together to generate a cumulative risk index or metric.

The CR model is premised on the observation that risk exposures lead to more adverse outcomes depending on the *number* of different risks experienced, rather than the nature of those risks, for instance duration or severity of risk exposure. Consequently, a drawback of this approach is that each individual risk factor contributes the same weight toward the CR index. Nevertheless, the cumulative effect of experiencing multiple risk factors is associated with increased risk for adverse outcomes (Rutter, 1978).

However, my interest is in the magnitude of the effect of individual putative risk factors as this could further inform prioritisation of need and guide on-going management from the point of referral in the stuttering population.

In order to estimate the individual (non-aggregated) effect of putative risk factors on internalising problems in adolescents who stutter, I first fitted univariate logistic regression models. There were five outcome measures in total, including SDQ Emotion Subscale parent-report and SMFQ (self-report) at age 14; SDQ Emotion Subscale self-report, Kessler-6 self-report, and diagnosis of anxiety or depression by a doctor at age 17. Scores on the mental health measures were dichotomised according to the clinical cut-off thresholds provided in the manual.

Five multiple logistic regression models were then fitted to test the additive effect of individual risk factors on each outcome variable separately, while taking into account the effect of other predictors. All predictor variables were binary and were selected based on the extensive literature pertaining to risk factors for adverse mental health, including SES (above or below the OECD 60% median poverty indicator), child sex (male/female), diagnosis of autism (yes/no), ADHD diagnosis (yes/no), parent mental health (mother or father ever diagnosed with anxiety or depression), bullying (ever bullied) and co-occurring speech and language problem (yes/no). All logistic regression models were fitted using the *survey* and *mitools* packages in R in order to pool results from the five imputed datasets.

Results

The final sample comprised all those participants who contributed data for the two binary predictor variables (stuttering variable and speech & language problems variable, $n = 17,256$). Of this sample, 565 cohort members reported stuttering, and 3,140 reported speech and language problems at least once during sweeps two and seven. Of the 565 cohort members in the stuttering group, 269 had co-occurring speech and language problems at some point in development (stutter plus speech & language problem group). Those cohort members who did not report stuttering or speech and language problems at any time point were assigned to the 'no difficulties' group, comprising 13,820 cohort members.

Table 15 provides the weighted descriptive statistics for all mental health measures (outcome variables). Participant characteristics by group and descriptive statistics for the original (unimputed) data can be found in Appendix G.

Table 15: Weighted descriptive statistics for all mental health measures by group

Group	Stuttering only group (n = 296)		Speech & Language Problems Group (n = 2,871)		Stutter and Speech & Language Problems Group (n = 269)		No Difficulties Group (n = 13,820)	
	Mean (SE)	95% CI	Mean (SE)	95% CI	Mean (SE)	95% CI	Mean (SE)	95% CI
SDQ Internalising Scores								
MCS2 Internalising Scores	3.04 (0.18)	2.68 – 3.40	3.56 (0.07)	3.43 – 3.70	3.75 (0.21)	3.32 – 4.18	2.65 (0.02)	2.61 – 2.70
MCS3 Internalising Scores	2.76 (0.18)	2.41 – 3.11	3.35 (0.07)	3.21 – 3.49	3.89 (0.25)	3.40 – 4.39	2.25 (0.02)	2.20 – 2.29
MCS4 Internalising Scores	3.01 (0.21)	2.60 – 3.42	3.82 (0.08)	3.67 – 3.98	4.32 (0.29)	3.75 – 4.89	2.46 (0.03)	2.40 – 2.53
MCS5 Internalising Scores	3.37 (0.24)	2.90 – 3.84	4.61 (0.11)	4.40 – 4.83	5.28 (0.30)	4.68 – 5.87	2.80 (0.03)	2.74 – 2.86
MCS6 Internalising Scores	3.83 (0.28)	3.26 – 4.39	5.15 (0.10)	4.95 – 5.34	5.89 (0.35)	5.19 – 6.60	3.32 (0.05)	3.22 – 3.43
MCS7 Internalising Scores	3.75 (0.30)	3.13 – 4.36	4.96 (0.12)	4.71 – 5.21	5.59 (0.37)	4.83 – 6.36	3.50 (0.04)	3.41 – 3.59
MCS Self-Report Measures								
SMFQ (MCS6)	5.97 (0.44)	5.10 – 6.84	5.70 (0.14)	5.43 – 5.97	5.53 (0.45)	4.64 – 6.41	5.48 (0.09)	5.29 – 5.67
Kessler-6 (MCS7)	6.98 (0.36)	6.27 – 7.69	7.28 (0.14)	6.99 – 7.56	7.02 (0.39)	6.25 – 7.78	7.17 (0.07)	7.03 – 7.31
SDQ Internalising score (MCS7)	5.74 (0.26)	5.22 – 6.27	5.98 (0.10)	5.79 – 6.18	5.78 (0.27)	5.25 – 6.32	5.55 (0.04)	5.46 – 5.63

Hypothesis 1: Symptom Trajectories

Four latent growth curve models were fitted to the multiply imputed dataset. The model intercept in these models represents scores at MCS2 (age 3) and the slope is measured in years corresponding to each sweep (age 3, 5, 7,

11, 14 and 17). Model 1 was the baseline model, which modelled the intercept and slope of internalising scores for the whole sample. Model 2 included the two binary grouping predictors to evaluate the effect of stuttering and speech and language problems on internalising scores at MCS2 and over time. Output from models one and two can be found in Appendix H. In order to model non-linear change over time, a quadratic term was added to Model 2 (Model 3). Model four included time-varying (mother and father Kessler scores at each time point) and time invariant covariates (child sex, child ethnicity, verbal ability at age 3, ASD or ADHD diagnosis, and SES (maternal education, OECD income quintiles and OECD 60% median poverty indicator) to examine whether symptom trajectories were robust to the addition of covariates. Model three (unadjusted) and model four (adjusted) are reported here (Table 16).

Unadjusted Model

After controlling for speech and language problems, there was a marginal effect of stuttering on the intercept ($b = 0.33$, $SE = 0.16$, $p = .047$) but not on the slope ($b = 0.08$, $SE = 0.05$, $p = .09$), suggesting that cohort members who reported stuttering on average scored 0.33 points higher at age 3 compared to non-stuttering peers, but did not differ in the rate of change in scores over time.

There was also an effect of speech and language difficulties on symptom trajectories, whereby cohort members who had ever reported speech and language problems scored, on average, 0.81 points higher at MCS2 relative to those who had not reported speech and language problems, with symptom scores increasing by 0.20 points for every sweep. There was also evidence of a non-linear relationship between speech and language problems and internalising scores, suggesting that the rate of increase in scores decreases as time increases ($b^2 = -0.011$, $SE = .002$, $p < .001$).

To check whether there was an interaction between stuttering and speech and language problems, an interaction term was added to the model, but I found no evidence of an interaction for the intercept ($b = -0.13$, $SE = 0.328$, p

= .69) nor the linear ($b = 0.028$, $SE = 0.096$, $p = .78$) or quadratic ($b^2 = 0.001$, $SE = 0.01$, $p = .92$) slopes. This would suggest that the additive effect of having both a stutter and speech and language problems is associated with the highest internalising scores at age 3. As can be seen in Figure 7, cohort members with both a stutter and speech and language problems had the highest scores, followed by the speech and language problems only group. The stuttering and no difficulties groups show the greatest differences in internalising scores in the first few sweeps; cohort members with no difficulties had lower scores to begin with but reached a similar point as the stuttering group by the final sweep.

Adjusted Model

After adjusting for child sex, ethnic group, SES, co-occurring autism or ADHD, age 3 verbal ability and parent mental health, there was no longer an effect of stuttering on age three internalising scores ($b = 0.19$, $SE = 0.16$, $p = .26$) nor an effect of stuttering on rate of change in scores over time ($b = 0.05$, $SE = 0.05$, $p = .27$). The addition of these covariates, however, did not change the significance of the effects of speech and language problems on internalising symptom trajectories. Following adjustment, cohort members with speech and language problems scored, on average, 0.39 points higher at age 3 than those without speech and language problems, and scores increased by 0.1 points per sweep. Thus, after accounting for co-occurring speech and language problems and other potential confounding factors, stuttering is not associated with internalising symptoms.

Table 16: Regression analyses for the unadjusted and adjusted latent growth curve models with stuttering and speech and language problems predictor variables

Predictor	Parameter	Unadjusted model				Adjusted model			
		Coefficient	SE	df	p-value	Coefficient	SE	df	p-value
Stutter	Intercept	0.33	0.16	2020.75	.047	0.19	0.16	805.702	.26
	Linear Slope	0.08	0.05	73.66	.09	0.05	0.05	62.783	.27
	Quadratic Slope ²	-0.005	0.003	18.09	.10	-0.003	0.003	18.25	.29
Speech & language problems	Intercept	0.81	0.07	119.65	<.001	0.39	0.072	80.977	<.001
	Linear Slope	0.20	0.02	88.57	<.001	0.10	0.023	66.530	<.001
	Quadratic Slope ²	-0.011	0.002	35.47	<.001	-0.006	0.002	39.498	.001

Note – model fit indices for unadjusted model (CFI = 0.97, TLI = 0.95, RMSEA = 0.04, SRMR = 0.03); model fit indices for adjusted model (CFI = 0.99, TLI = 0.98, RMSEA = 0.011, SRMR = 0.03)

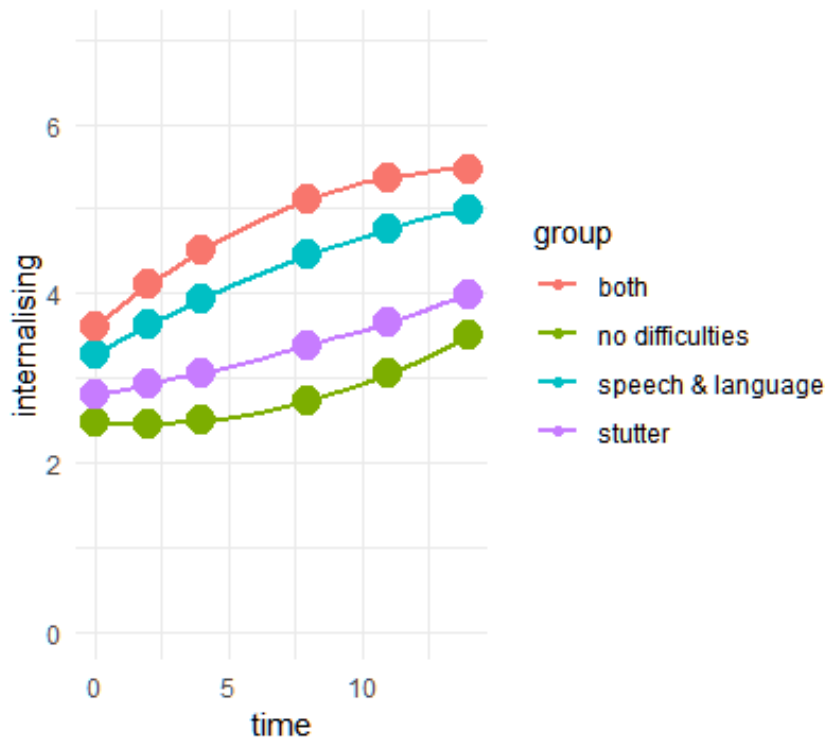


Figure 7: Internalising symptom trajectories for all four groups from MCS2 to MCS7 (unadjusted model). y-axis shows mean internalising symptom scores, x-axis displays time (year) from MCS2 (first data point).

Hypothesis 2: Stuttering and Self-harm

Table 17 shows the proportion of cohort members in each group who reported a diagnosis of anxiety or depression at age 17, self-harm at ages 14 and 17, and suicide attempt at 17.

Table 17: Summary statistics for self-report measures at age 14 and 17. Frequency table shows *n* participants and the corresponding proportion (%) of the group who responded 'yes'.

Variable	Stutter only (n=296)	Speech/language problem only (n=2,871)	Stutter & speech/language problems (n=269)	No reported problems (n=13,820)
Diagnosis of serious anxiety or depression	25 (8.45%)	273 (9.51%)	22 (8.18%)	955 (6.91%)
Self-harm (age 14)	35 (11.82%)	340 (11.84%)	25 (9.29%)	1529 (11.06%)
Self-harm (age 17)	52 (17.57%)	551 (19.19%)	57 (21.19%)	2446 (17.70%)
Suicide attempt	17 (5.74%)	175 (6.10%)	14 (5.20%)	687 (4.97%)

A total of 3,106 cohort members in the whole sample reported deliberate self-harm at age 17. In the unadjusted model, in which stuttering was the only predictor, there was no significant association between having a stutter and odds of disclosing self-harm at age 17 (OR=1.01, 95% CI[0.76, 1.34], $p = .96$). To determine whether any other factors may be masking an association between stuttering and self-harm, the adjusted model has been reported (Table 18). After adjusting for other covariates, there was no evidence of an effect of stuttering on likelihood of reporting self-harm at age 17 years (OR = 1.04, 95% CI[0.78, 1.40], $p = .78$).

Table 18: Results of weighted multiple logistic regression model, adjusting for other factors in the association between stuttering and odds of reporting self-harm at age 17 years

Predictor	OR [95% CI]	df	p-value
(Intercept)	0.12 [0.09, 0.16]	15.58	<.001
Stutter (yes)	1.04 [0.78, 1.40]	41.99	.78
Speech & Language problems (yes)	1.09 [0.95, 1.26]	41.16	.22
Child sex (female)	1.99 [1.80, 2.21]	73.91	<.001
OECD second income quintile	0.99 [0.84, 1.17]	43.29	.94
OECD third income quintile	0.95 [0.76, 1.18]	15.06	.60
OECD fourth income quintile	0.97 [0.78, 1.20]	17.51	.76
OECD highest income quintile	0.96 [0.80, 1.15]	93.89	.66
Mother NVQ equivalent Level 2	1.13 [0.86, 1.48]	8.79	.33
Mother NVQ equivalent Level 3	1.34 [0.91, 1.97]	7.79	.12
Mother NVQ equivalent Level 4	1.31 [1.00, 1.71]	11.15	.05
Mother NVQ equivalent Level 5	1.55 [1.07, 2.23]	19.03	.02
Mother NVQ equivalent overseas	1.08 [0.72, 1.62]	13.30	.71
Mother Kessler Total score (mcs7)	1.05 [1.03, 1.06]	11.72	<.001
Father Kessler Total score (mcs7)	1.02 [1.01, 1.04]	18.15	.01
Mother ever diagnosed with anxiety or depression (yes)	1.20 [1.08, 1.33]	122.79	.001

Father ever diagnosed with anxiety or depression (yes)	1.15 [1.01, 1.31]	56.04	.04
Child ethnicity (black)	0.66 [0.44, 0.98]	19.03	.04
Child ethnicity (Indian)	0.69 [0.45, 1.05]	35.49	.08
Child ethnicity (Mixed)	0.89 [0.69, 1.16]	392.14	.39
Child ethnicity (Pakistani & Bangladeshi)	0.52 [0.37, 0.72]	16.72	.001
Child ethnicity (other)	0.91 [0.55, 1.51]	35.16	.70

Note: reference categories for variables with more than two factors were NVQ Level 1 (lowest qualification); OECD lowest income quintile; and white ethnicity.

Hypothesis 3: Stuttering and clinical-level symptoms

Approximately 7% of cohort members in the whole sample reported that they had been diagnosed with serious anxiety or depression by a doctor ($n = 1, 275$). Although stuttering was not found to be associated with an anxiety or depression diagnosis at MCS7 in the unadjusted model in which stuttering was the only predictor (OR = 1.07, 95% CI[0.73, 1.57], $p = .73$), I again considered whether any other factors may be masking the effect of stuttering. Results of the adjusted model indicate no effect of stuttering on the odds of being diagnosed with anxiety or depression (OR = 1.01, 95% CI [0.69, 1.47], $p = .97$), even after accounting for a number of other factors (Table 19).

Table 19: Weighted multiple logistic regression model, adjusting for other factors in association between stuttering and odds of being diagnosed with anxiety or depression at 17.

Predictor	OR [95% CI]	df	p-value
(Intercept)	0.04 [0.03, 0.05]	24.33	<.001
Stutter (yes)	1.01 [0.69, 1.47]	65.93	.97
Speech & Language Problems (yes)	1.30 [1.02, 1.65]	13.72	.04
Child sex (female)	2.51 [2.19, 2.88]	160.65	<.001
OECD second income quintile	0.92 [0.77, 1.10]	877.22	.36
OECD third income quintile	0.98 [0.76, 1.26]	24.01	.85
OECD fourth income quintile	0.96 [0.74, 1.26]	25.80	.78

OECD highest income quintile	0.83 [0.64, 1.08]	75.83	.17
Mother NVQ equivalent Level 2	0.91 [0.69, 1.19]	14.33	.47
Mother NVQ equivalent Level 3	0.83 [0.63, 1.10]	95.40	.19
Mother NVQ equivalent Level 4	0.98 [0.76, 1.25]	60.09	.86
Mother NVQ equivalent Level 5	0.94 [0.57, 1.58]	23.00	.82
Mother NVQ equivalent overseas	0.65 [0.27, 1.56]	7.45	.28
Mother's Kessler total score (MCS7)	1.06 [1.04, 1.08]	14.89	<.001
Father's Kessler total score (MCS7)	1.03 [1.01, 1.05]	21.95	.01
Mother ever diagnosed with anxiety or depression (yes)	1.93 [1.61, 2.32]	22.58	<.001
Father ever diagnosed with anxiety or depression (yes)	1.21 [1.03, 1.43]	65.76	.02
Child ethnicity (black)	0.67 [0.39, 1.16]	39.85	.15
Child ethnicity (Indian)	0.64 [0.36, 1.15]	133.31	.14
Child ethnicity (Mixed)	0.81 [0.53, 1.24]	56.51	.33
Child ethnicity (Pakistani & Bangladeshi)	0.37 [0.23, 0.60]	20.04	<.001
Child ethnicity (other)	0.33 [0.08, 1.36]	11.07	.11

Note: reference categories for variables with more than two factors were NVQ Level 1 (lowest qualification); OECD lowest income quintile; and white ethnicity.

Hypothesis 4: multiple risk factor models for stuttering group

In order to estimate the extent to which certain factors may affect risk of clinical-level symptoms in the stuttering group ($n = 565$), univariate and multiple logistic regression analyses were performed. Scores on four mental health measures (SDQ Emotion subscale-parent report, SDQ Emotion subscale self-report, SMFQ, and Kessler-6) were dichotomised and entered as binary variables (above or below threshold) for each analysis. The final model includes diagnosis of anxiety or depression at age 17 as a binary outcome variable.

Univariate logistic regression models

The size of the effect of each predictor on likelihood of clinical-level symptoms was examined through a series of univariate analyses (see Appendix I). The independent effect of eight binary predictor variables on odds of scoring above threshold on the four mental health measures and anxiety or depression diagnosis is reported.

Parent-report measures

SDQ Emotion Subscale: Increased odds of parents reporting above-threshold scores on the SDQ Emotion Subscale were associated with both child and family factors. Cohort members who came from lower SES backgrounds (OR = 2.36, 95% CI [1.34, 4.13], $p = .003$) and whose mother had ever been diagnosed with anxiety or depression (OR = 2.35, 95% CI [1.34, 4.13], $p = .003$) had greater odds of clinical-level scores on this subscale. Cohort members who were female (OR = 1.82, 95% CI [1.09, 3.04], $p = .02$), had ever reported speech and language problems (OR = 2.09, 95% CI [1.07, 4.06], $p = .03$) or had a diagnosis of Autism (OR = 5.62, 95% CI [2.79, 11.33], $p < .001$) or ADHD (OR = 3.30, 95% CI [1.37, 7.97], $p = .01$) had increased odds of scoring above threshold on this measure.

Self-report measures

Kessler-6: Results of the univariate models indicated that female sex (OR = 3.41, 95% CI [1.71, 6.81], $p = .001$), having a mother (OR = 3.08, 95% CI [1.58, 6.01], $p = .001$) or father (OR = 1.96, 95% CI = [1.03, 3.75], $p = .04$) who had ever been diagnosed with serious anxiety or depression, and lower SES (OR = 2.00, 95% CI [1.10, 3.67], $p = .02$) significantly predicted likelihood of scoring above threshold on the Kessler-6 in the stuttering group.

Short Moods & Feelings Questionnaire: Scoring above clinical threshold on the SMFQ was associated with female sex (OR = 4.20, 95% CI [2.23, 7.88], $p < .001$) and lower SES (OR = 1.87, 95% CI [1.02, 3.41], $p = .04$).

SDQ Emotion Subscale: Female sex (OR = 5.80, 95% CI [2.98, 11.30], $p = .001$) and having a mother who had ever been diagnosed with serious

anxiety or depression (OR = 2.75, 95% CI [1.37, 5.50], $p = .005$) were associated with above threshold scores on the emotion subscale at age 17.

Anxiety or depression diagnosis: Reporting a diagnosis of serious anxiety or depression at age 17 was associated with being female (OR = 2.78, 95% CI [1.26, 6.12], $p = .01$) and having a mother with a diagnosis of anxiety or depression (OR = 2.50, 95% CI [1.17, 5.31], $p = .02$).

In summary, several factors were found to be associated with scores that exceeded clinical threshold, as well as with the likelihood of being diagnosed with anxiety or depression, among adolescents who stutter. A comparison of the size of these non-amalgamated effects can be found in Appendix I.

Multiple Logistic Regression Models

In order to test the additive effect of these risk factors, while accounting for the effect of other factors, multiple logistic regression models were fitted for each mental health measure. Results of the five multiple regression models indicated that female sex was consistently associated with increased odds of clinical-level symptoms on all measures in the stuttering sample (Table 20). In addition, at age 17, having a mother who had ever reported a diagnosis of anxiety or depression increased odds of scoring above threshold on the Kessler-6 (OR = 2.31, 95% CI [1.13, 4.71], $p = .02$) and the SDQ Emotion subscale (OR = 2.56, 95% CI [1.20, 5.45], $p = .01$), both of which are self-report measures.

For the parent-reported SDQ Emotion subscale, cohort members who were female (OR = 2.46, 95% CI [1.38, 4.38], $p = .002$) or who had a diagnosis of autism (OR = 5.37, 95% CI [2.17, 13.29], $p < .001$) had increased odds of obtaining clinical-level scores. However, the wide confidence interval for autism diagnosis should be noted. Table 20 presents the odds ratios associated with each predictor variable for the five outcome measures.

Figures 8 – 12 visualise the size of the effect (odds ratio) associated with each predictor in the five multiple logistic regression models.

Table 20: Results of five multiple logistic regression models, examining the effect of predictors on likelihood of scoring above threshold on the SDQ, SMFQ and Kessler-6, and being diagnosed with anxiety or depression at 17 in the stuttering group (n = 565).

	Predictor	OR [95% CI]	df	p-value
SDQ Emotion subscale (parent-report) at 14	(Intercept)	0.05 [0.01, 0.19]	12.74	<.001
	Female sex	2.46 [1.38, 4.38]	447.91	.002
	Below 60% OECD poverty indicator	1.82 [1.00, 3.33]	112.16	.05
	Mother ever diagnosed with anxiety or depression	1.78 [1.00, 3.16]	196.48	.05
	Father ever diagnosed with anxiety or depression	1.04 [0.51, 2.11]	43.10	.91
	ASD diagnosis	5.37 [2.17, 13.29]	236.84	<.001
	ADHD diagnosis	1.22 [0.40, 3.77]	47.97	.72
	Co-occurring speech & language problems	1.48 [0.70, 3.13]	22.67	.29
	Ever bullied	1.10 [0.36, 3.32]	10.78	.85
	(Intercept)	0.02 [0.00, 0.11]	14.58	<.001
SMFQ (self-report) at age 14	Female sex	4.48 [2.31, 8.71]	86.69	<.001
	Below 60% OECD poverty indicator	1.51 [0.78, 2.93]	66.32	.22
	Mother ever diagnosed with anxiety or depression	1.20 [0.59, 2.46]	48.35	.61
	Father ever diagnosed with anxiety or depression	1.30 [0.50, 3.41]	13.87	.56
	ASD diagnosis	1.46 [0.34, 6.16]	20.88	.59

	ADHD diagnosis	0.96 [0.24, 3.89]	24.22	.95
	Co-occurring speech & language problems	0.94 [0.49, 1.81]	229.87	.86
	Ever bullied	4.36 [0.81, 23.41]	10.34	.08
SDQ Emotion Subscale (self- report) at 17	(Intercept)	0.02 [0.00, 0.07]	45.49	<.001
	Female sex	6.16 [3.00, 12.65]	228.43	<.001
	Below 60% OECD poverty indicator	1.09 [0.47, 2.50]	27.03	.83
	Mother ever diagnosed with anxiety or depression	2.56 [1.20, 5.45]	165.38	.01
	Father ever diagnosed with anxiety or depression	0.81 [0.29, 2.26]	17.57	.67
	ASD diagnosis	3.04 [0.79, 11.70]	31.75	.10
	ADHD diagnosis	0.63 [0.07, 5.64]	15.09	.66
	Co-occurring speech & language problems	0.64 [0.28, 1.43]	66.00	.27
	Ever bullied	2.37 [0.59, 9.45]	15.47	.20
Kessler-6 (self- report) at age 17	(Intercept)	0.01 [0.003, 0.06]	31.38	<.001
	Female sex	3.94 [1.85, 8.39]	49.66	.001
	Below 60% OECD median poverty indicator	1.39 [0.72, 2.66]	113.45	.32
	Mother ever diagnosed with anxiety or depression	2.31 [1.13, 4.71]	287.23	.02
	Father ever diagnosed with anxiety or depression	1.58 [0.78, 3.20]	130.29	.21
	ASD diagnosis	2.18 [0.57, 8.35]	23.46	.24
	ADHD diagnosis	0.97 [0.23, 4.08]	23.35	.97

	Co-occurring speech & language problems	1.10 [0.51, 2.40]	42.33	.80
	Ever bullied	3.57 [0.90, 14.15]	21.33	.07
Diagnosed with anxiety or depression (age 17)	(Intercept)	0.01 [0.001, 0.07]	14.57	<.001
	Female sex	3.02 [1.37, 6.65]	46.10	.01
	Below 60% OECD poverty indicator	1.25 [0.61, 2.57]	126.07	.54
	Mother ever diagnosed with anxiety or depression	1.89 [0.83, 4.29]	66.48	.13
	Father ever diagnosed with anxiety or depression	1.60 [0.63, 4.07]	21.04	.31
	ASD diagnosis	1.66 [0.32, 8.75]	29.00	.54
	ADHD diagnosis	1.00 [0.18, 5.45]	41.97	1.00
	Co-occurring speech & language problems	1.05 [0.43, 2.55]	35.51	.92
	Ever bullied	4.75 [0.61, 36.93]	10.42	.12

Age 14 mental health measures

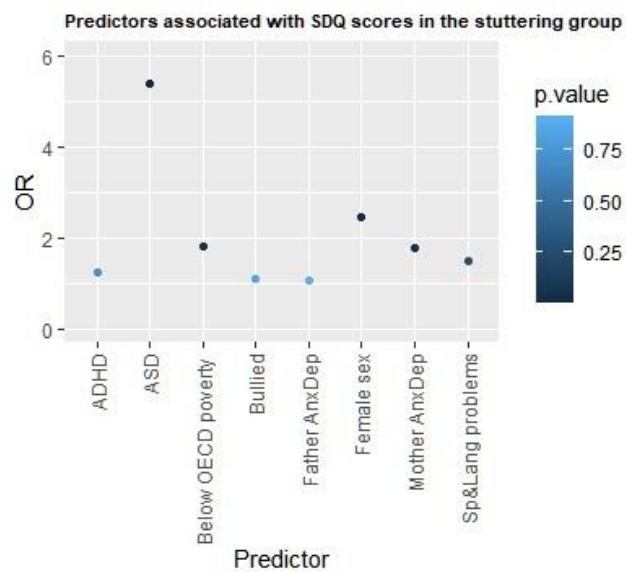


Figure 8: Plot showing effect of predictors on odds of scoring above threshold on the parent-reported SDQ emotion subscale (n = 565)

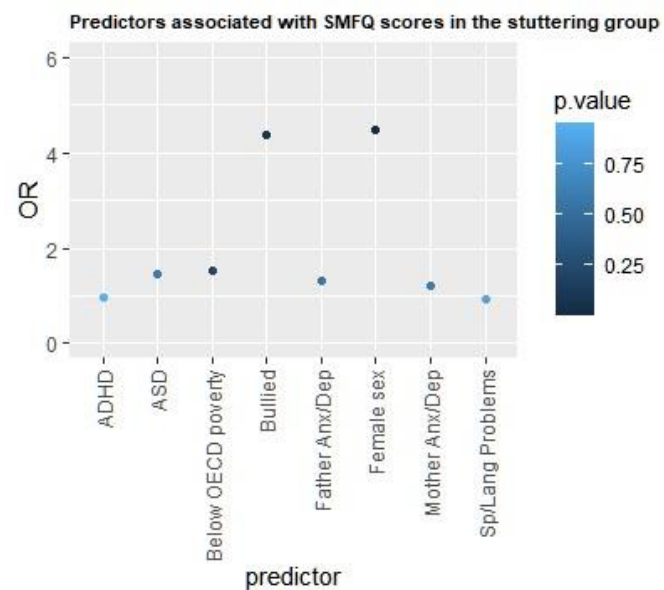


Figure 9: Plot showing effect of predictors on odds of scoring above threshold on the SMFQ (n = 565)

Age 17 mental health measures

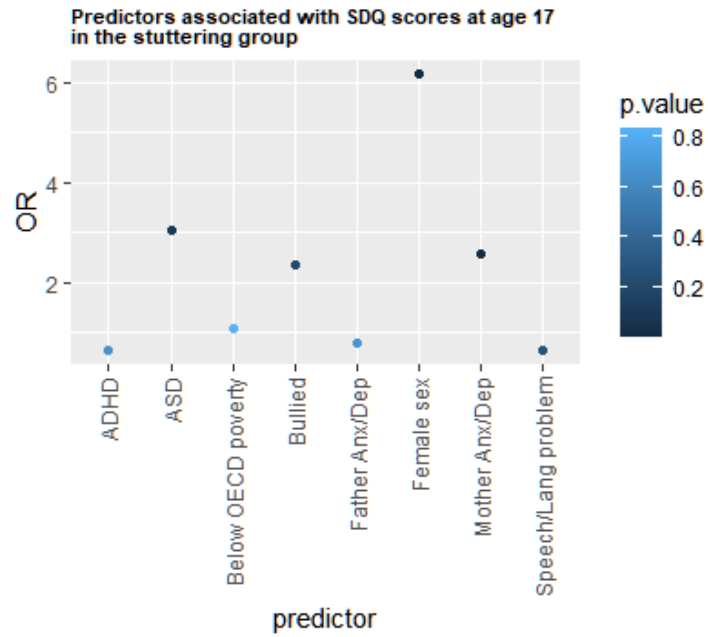


Figure 10: Plot showing effect of predictors on odds of scoring above threshold on the SDQ Emotion subscale self-report ($n = 565$)

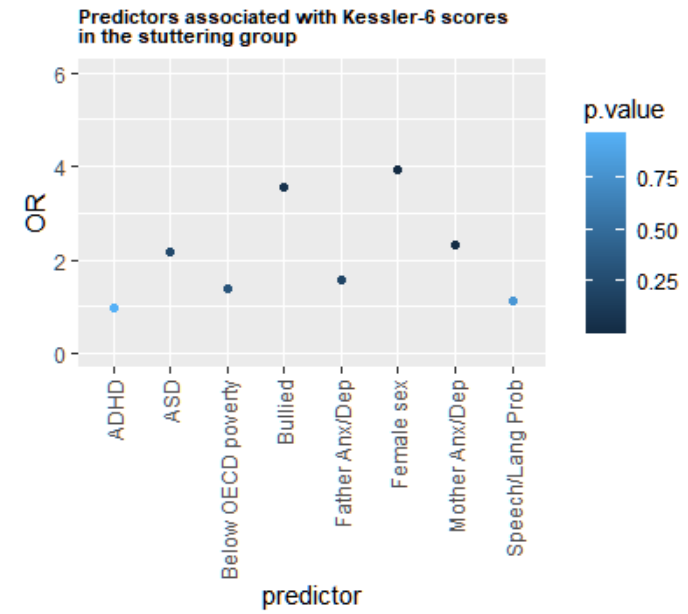


Figure 11: Plot showing effect of predictors on odds of scoring above threshold on the Kessler-6 ($n = 565$)

Age 17 mental health measures continued.

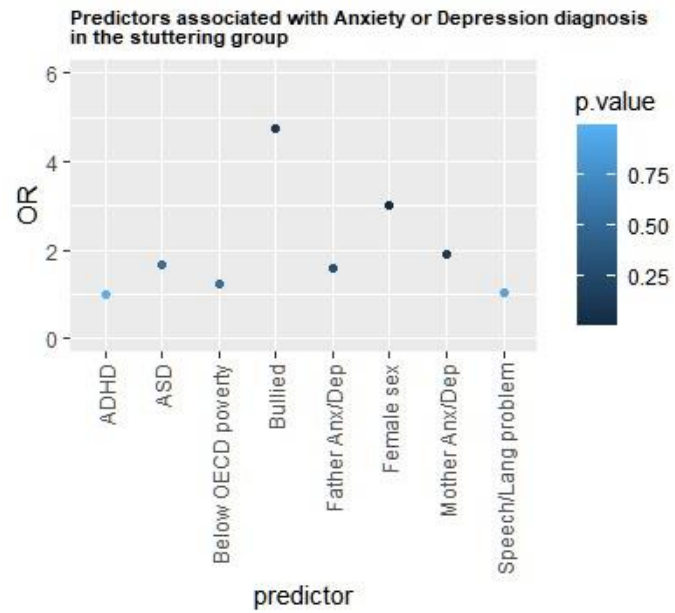


Figure 12: Plot showing effect of predictors on odds of a serious anxiety or depression diagnosis at 17 (n = 565)

Discussion

This study investigated the effect of stuttering on internalising symptom trajectories in children and adolescents, taking account of co-occurring speech and language problems. Additionally, the study examined cumulative risks that may elevate anxiety and depression symptoms in adolescents who stutter.

Internalising symptoms trajectories

The first hypothesis asked whether children and adolescents who stutter (aged 3 – 17 years) report more internalising symptoms with age than children who do not stutter. Latent growth curve models showed no effect of stuttering on symptom trajectories after adjustment for other child- and family- factors. This aligns with previous population studies, which have found no differences in anxiety symptoms in community samples (Andrews & Harris, 1964; Smith et al., 2017).

However, there was an effect of speech and language problems even after adjustment for covariates; cohort members reported to have speech and language problems during development had higher internalising scores at age 3 and steeper symptom trajectories over time than children without speech and language problems.

In both the unadjusted and adjusted models, cohort members with both stuttering and speech and language problems had the highest internalising scores at age 3, with an increasing symptom trajectory over time. This underscores the additive effect of co-occurring communication difficulties on internalising symptoms in children who stutter.

The SDQ is a parent-report measure and therefore the effect of co-occurring communication difficulties may exacerbate the concern parents already have on account of their child stuttering. Alternatively, the challenges these children may face in terms of interacting with peers and unfamiliar

environments may explain the increasing symptom scores among these children. The present finding supports previous studies that have shown that children who stutter who have co-occurring communication difficulties report higher anxiety symptoms than children who stutter without additional needs (Blood et al., 2007; Briley, O'Brien & Ellis, 2019).

Nevertheless, the present study results differ from the findings of McAllister (2016) who analysed cross-sectional data from three sweeps of the MCS to examine the social, emotional and behavioural development of children who stutter compared to typically developing peers. After adjusting for sex, verbal and non-verbal abilities, maternal education and family socio-economic status, McAllister found that children who stutter showed significantly higher scores on all subscales of the SDQ at ages five and eleven years. At age three, adjusted analyses indicated groups differed only in the total difficulties score. There are several differences between the present study and that of McAllister (2016), which likely contribute to the differing findings.

As McAllister acknowledges, the children in each sweep were not necessarily the same children, whereas the present study plotted symptom trajectories for the same cohort members to examine within-person change over time. This also means that some individuals in the stuttering group may have resolved their stutter at later time points. While this study examined internalising symptoms, as calculated by summing the Emotion and Peer Problem subscales of the SDQ, McAllister compared scores on each of the SDQ subscales. In the McAllister (2016) study, cohort members were divided into two groups based on the same question used in the present study (parental concern about speech and language development). Although the typically developing group comprised cohort members who had no reported speech difficulties at each age point, the stuttering group may have included cohort members who also had other parent-reported communication difficulties. Therefore, it may be that the inclusion of children who stutter with co-occurring speech, language and communication difficulties contributed to the differences in symptom scores observed in the stuttering group relative to the non-stuttering group in this earlier study.

Stuttering and self-harm

The second hypothesis considered whether there would be differences in the proportion of adolescents in the stuttering and non-stuttering group reporting deliberate self-harm. After controlling for co-occurring speech and language problems and other confounding factors, there was no evidence that the proportion of cohort members reporting self-harm at age 17 differed from those who do not stutter. This is consistent with findings from another UK population study, which failed to find any association between stuttering at age eight years and self-harm at age 16 (McAllister et al., 2023).

The existing literature points to a number of genetic, biological, psychiatric, psychological, social and cultural factors associated with increased risk for deliberate self-harm in adolescence (Hawton, Saunders & O'Connor, 2012). For instance, increased likelihood of deliberate self-harm has been associated with female sex, low parental socio-economic status, bullying victimisation, parental mental health disorder and adverse childhood experiences (Hawton, Saunders & O'Connor, 2012; Lodebo et al., 2017; Page et al., 2014). In the present study, female sex and parental mental health were found to significantly predict self-harm in the sample. Interestingly, higher SES (maternal education indicator) was associated with increased odds for self-harm in the present study, even though low SES has previously been associated with self-harm.

Stuttering and diagnosis of anxiety and depression

The third hypothesis was concerned with whether adolescents who stutter differ from non-stuttering peers in terms of likelihood of reporting anxiety or depression diagnosed by a health professional at age 17. There was no evidence of an association between stuttering and odds of being diagnosed with anxiety or depression. There was evidence of an effect of speech and language problems on odds of anxiety or depression diagnosis, which would complement the existing literature showing elevated risk for anxiety among children with language difficulties (Curtis et al., 2018; Forrest et al., 2020;

Hentges et al., 2021; Yew & O’Kearney, 2013). An earlier study using data from another British birth cohort, the National Child Development Study (NCDS), compared scores indicative of mental health disorders in a group of adolescents who stutter, a group who report other speech difficulties and adolescents with no difficulties (McAllister, Collier & Shepstone, 2013). After accounting for confounding factors, other speech difficulties were found to be associated with scores indicative of increased risk for mental health disorders on the Rutter Malaise Inventory relative to controls. However, stuttering was not associated with increased risk for mental health disorders in this study. It should be noted that the McAllister et al. (2013) study compared stuttering with ‘other *speech* difficulties’, whereas the speech and language problem group in the present study comprised cohort members with a broader range of speech and language difficulties.

In this study, diagnosis by a health professional was considered evidence of clinical anxiety or depression in so far as symptoms were recognised as impacting everyday functioning. Higher rates of clinical anxiety disorders in children and adolescents who stutter have been reported in previous studies (Iverach et al., 2016; Gunn et al., 2014). However, these studies were not community samples. Therefore, it may have been that these studies overestimated rates of clinical anxiety due to the fact that families accessing services may be in greater need of support and thus more likely to present with clinical-level symptoms. Equally, relying on self-report of diagnosis introduces selection bias as cohort members would have needed to attend an appointment to be diagnosed by a GP and many adolescents with mental health problems, particularly emotional disorders, do not access services (Radez et al., 2021). Consequently, I may have underestimated rates of anxiety and depression in the population.

Factors associated with risk for anxiety or depression

The fourth and final hypothesis considered whether multiple risk factor models could contribute to our understanding of the heterogeneity in internalising symptom reporting among adolescents who stutter. In a large

cohort of adolescents with a history of stuttering, the multiple risk models show that female sex is robustly associated with above-threshold internalising symptom scores and clinical diagnosis of anxiety or depression. Maternal mental health was associated with increased odds of psychological distress.

Autism spectrum conditions were only associated with emotional symptom scores that exceeded clinical threshold as reported by parents. The SDQ was the only outcome variable relying on parent-report included in the multiple risk models, which may have influenced present findings. For example, parents are likely to be concerned about their child's development and future aspirations if their child is not developing at a similar rate to their peers. This could result in parents reporting more symptoms because they are concerned about their child and the challenges they may face. Differences in symptom reporting by respondent is well documented (see DeLos Reyes & Kazdin, 2005) but self-report data were not available for this measure at age 14, preventing comparisons with parent-reported scores.

It is somewhat surprising that parent mental health was not consistently associated with odds of scoring above clinical cut-off on included mental health measures, nor with diagnosis of anxiety or depression in adolescents who stutter. Parental anxiety and depression have been cited among the strongest predictors of these conditions in children and adolescents in the general population (Thapar et al., 2012; Hyland et al., 2016). In chapter four, I report that family history of adverse mental health significantly predicted anxiety and depression scores in a sample of school-aged children who stutter. However, a number of risk and protective factors likely play a role in development of anxiety and depression, and therefore the interplay between a number of factors may help to explain the null effect once all predictors were added into the model.

There are both theoretical and clinical implications of this finding. Firstly, it will be important to consider presence of these risk factors in assessment and clinical management of children and adolescents who stutter to promote

the psychosocial well-being of these individuals. Secondly, future research should take such putative risk factors into account when comparing symptoms among children and adolescents who stutter and their non-stuttering peers. While it has not been possible to estimate the effect of other factors specific to stuttering, such as stuttering severity, age of onset, and persistence, on risk for anxiety or depression in this population, it is important that these putative risk factors for mental health are also considered to accurately estimate prevalence of internalising problems relative to the general population.

Clinical and sub-clinical symptomology

A question of interest, and clinical importance, is whether elevated scores on mental health measures reflect clinical-level symptomology in the stuttering population. This study did not compare the likelihood of scoring above clinical threshold on included mental health measures in cohort members who do and do not stutter. Nevertheless, self-report of anxiety or depression diagnosis and deliberate self-harm do offer insight into internalising problems that are of clinical concern. Based on the current findings, adolescents who stutter do not appear to be at greater risk of deliberate self-harm nor are they more likely to receive a diagnosis of anxiety or depression compared to those who do not stutter. This would perhaps suggest that stuttering can be associated with elevated anxiety and depression symptoms but not necessarily severe enough to warrant a clinical diagnosis in adolescence.

While the literature suggests adults who stutter are at increased risk of social anxiety disorder relative to the non-stuttering population (Blumgart Tran & Craig, 2010; Craig & Tran, 2014; Iverach & Rapee, 2014), research findings in children and adolescents are more mixed. Some earlier studies that have reported an association between stuttering and increased anxiety symptoms, have equally reported that symptom scores fell within the normative range (Blood et al., 2007; Gunn et al., 2014; Iverach et al., 2016; Iverach et al., 2017b).

Limitations

There are several limitations to the present study. Identification of stuttering among cohort members was dependent on parent-report rather than clinical assessment; therefore important information about the age of onset, severity and nature of the stutter is missing. Additionally, while the decision to allocate cohort members to the stuttering group based on report of stuttering at any time point increased sample size and mitigated the impact of subject attrition on analyses, it introduced within-participant variability. For instance, it was unclear which cohort members continued to stutter at age 17 and who recovered, which could affect mental health and well-being. Similarly, the characteristics of the speech and language problem group will be variable due to the fact that it is not possible to ascertain the nature, severity or duration of communication problems.

Clinical Implications

The findings from this study contribute further to the evidence base around the social and emotional development of children and young people who stutter. Although I found no evidence that stuttering on its own affected internalising symptom trajectories, cohort members who stuttered with co-occurring speech and language difficulties had the steepest trajectories, indicating a need to carefully manage these children and promote resilience. In fact, while cohort members reported to both stutter and have speech and language problems had the highest internalising scores, this group was followed by the speech and language problem only group, and then the stuttering group. The present study potentially offers further insight into the differing mental health profiles of children and adolescents with a range of communication difficulties.

While a number of risk factors for anxiety and depression have been identified in the general population, the present study contributes further to our understanding of risk factors involved in clinical-level symptoms in the stuttering population in particular. Prevalence and persistence of stuttering may be higher amongst boys than girls, but the present findings underscore

the need to be alert to and manage heightened risk for internalising problems among girls who stutter. Improving our understanding of factors associated with elevated risk for heightened symptomology in the stuttering population has the potential to aid clinicians in off-setting development of internalising problems through appropriate assessment and early identification. Holistic assessment of the child, family and environmental factors that could contribute towards internalising problems in adolescence offers an opportunity to work with all agencies to build resilience in those children likely to be at risk.

Conclusion

Longitudinal analysis of data obtained from the UK Millennium Cohort Study indicated that internalising symptom trajectories of children and adolescents who stutter do not differ from non-stuttering peers. However, individuals who stutter with co-occurring speech and language problems may be at risk for elevated internalising symptoms during development. Stuttering on its own is not associated with increased odds of self-harm or diagnosis of anxiety and depression in adolescence. However, similarly to the general population, being female and having a mother with anxiety or depression, are associated with increased odds of clinical-level symptoms in the stuttering population. Further longitudinal research is needed, which takes a broader number of factors into account when evaluating risk for internalising problems in young people who stutter compared to those who do not stutter.

Chapter 6

General Discussion

The aim of this thesis was to investigate whether children and adolescents who stutter are at risk of elevated anxiety and depression symptoms compared with their non-stuttering peers, which includes those with typical language and those with other speech/language difficulties. In particular, this research aimed to answer two questions:

- Are children and adolescents who stutter more likely to experience anxiety and/or depression symptoms than non-stuttering peers (those with typical language and those with other language/speech difficulties)?
- What other factors moderate the relationship between stuttering and anxiety and/or depression?

The original plan had been to conduct three studies that directly addressed these questions:

1. A systematic review of the extant literature that examined anxiety and depression symptoms in children (2 – 25 years) who did and did not stutter.
2. A national online questionnaire to be completed by children (8-13 years) who stutter and live in the UK. Scores on the mental health measure would be compared to population norms.
3. A comparative study, in which scores on mental health and bullying measures were compared in three cohorts of children (8-13years): children who stutter; children with language difficulties; children with typically developing speech and language skills. The plan had been to draw the non-stuttering groups from existing data obtained from the Surrey Communication & Language in Education Study (SCALES; Norbury, 2016).

Amended research plan

The systematic review and meta-analysis were completed as planned. Originally, the upper age limit was 25 years in order to reflect current conceptualisations of adolescence (Sawyer et al., 2018) and to maintain consistency with the UK Special Educational Needs Disability (SEND) Code of Practice in England (Children & Families Act, 2014). However, contrary to the protocol, the upper age limit was reduced to 18 years following recommendation by peer reviewers. The second study involved analysis of data obtained through an online questionnaire, however the pre-registered analysis plan needed to be amended in response to the pandemic and the potential impact on population mental health. The plan for the third study was re-formulated in response to the global coronavirus pandemic, which caused considerable issues with participant recruitment as well as recognised impacts on population mental health as a result of social distancing measures and the pandemic itself. In place of empirical data collection, secondary data were analysed from the Millennium Cohort Study for the final study.

Nevertheless, despite the challenges associated with the pandemic context, the association between internalising symptoms and speech-language status was examined in three pre-registered studies in an effort to address the original research questions. This chapter summarises the findings from each of these studies and considers the theoretical and clinical implications for children and young people who stutter and their families.

Summary of findings

Study 1: Systematic review

The first study systematically synthesised the existing literature comprising studies that had compared anxiety and depression symptom reporting in children and adolescents (3 – 18 years) who do and do not stutter. The results of the meta-analysis ($k=11$) indicated a moderate effect size difference between school-aged children (3 – 18 years) who stutter and their

non-stuttering peers in self- and parent-reported anxiety symptoms. The majority of included studies focused on school-aged children and adolescents (7 – 18 years). There was only one pre-school study that met criteria for inclusion in the review, highlighting the need for further research into very young children who stutter. Although there were insufficient data to quantitatively analyse depression symptoms in this age group, larger effect sizes were observed in studies comprising older participants.

A critical finding from the systematic review was the heterogeneity in symptom reporting in this clinical population, as highlighted by the varying effect sizes across included studies. While the aggregated effect size, which was based on synthesis of the eleven studies, indicated evidence of higher anxiety symptoms in the stuttering sample than the non-stuttering group, some included studies reported no significant between-group differences. Furthermore, qualitative examination of included studies highlighted issues in the extant literature, which may contribute to the variable results of previous studies. In particular, between-study variation relating to the construct and measurement of anxiety and the extent to which potential moderating factors were considered, such as co-occurring developmental difficulties and demographic factors associated with adverse mental health outcomes in the general population. Many studies did not report whether or how they had controlled for co-occurring communication and/or learning needs or existing mental health problems among participants. This is important given the substantial literature pointing to elevated risk of poor mental health and emotional outcomes among children with other neurodevelopmental conditions, including language and communication difficulties (Gnanavel et al., 2019; Livingston, Siegel & Ribary, 2018; Skokauskas & Gallagher, 2012; Yew & O’Kearney, 2013). This systematic review also found that methodological limitations previously identified in the field (see Menzies et al., 1999) continued to be observed in recent studies, in particular small sample sizes and reliance on clinical populations. Consequently, the conclusions drawn from the systematic review and meta-analysis are limited by the small number of included studies, small samples in included studies, and the variability in how anxiety was measured. Future research into

developmental stuttering needs to concentrate on larger community studies, which have the capacity to improve robustness of findings in this area and improve precision estimates.

To my knowledge, this is the first systematic review of anxiety and depression symptoms in children and adolescents who stutter and it highlighted the need for more robust research into the relationship between internalising symptoms and childhood stuttering. Nevertheless, the limitations of the review must also be acknowledged. The inclusion criteria stipulated that studies must involve a non-stuttering comparison group. While this permitted more direct comparison across studies, it led to exclusion of several studies, which therefore did not contribute towards the summary effect size. It should be recognised that the meta-analysis pooled anxiety scores from a variety of measures, and while efforts were made to try to group these measures in order to estimate the association between stuttering and social versus general anxiety, this grouping was subjective.

In summary, the findings of this review emphasised the variable outcomes of studies comparing anxiety symptoms in children who do and do not stutter, while also highlighting gaps in the literature around depression symptoms in this population and the role of factors that may moderate the association between stuttering and risk for poorer mental health outcomes in children and young people.

The two subsequent studies were designed to address some of the research gaps identified in the systematic review and enhance our understanding of the role of risk and protective factors in any association between stuttering and internalising symptoms.

Study 2: National online questionnaire study

Empirical data collected for the second project permitted analysis of anxiety and depression symptoms in a cohort of children who stutter recruited from a community sample in the UK. The pre-registered recruitment and analysis

plan for this study was amended prior to data analysis to mitigate the confound of the pandemic. Originally, children's scores on the Revised Children's Anxiety and Depression Scale (RCADS-25) were to be compared to the published population norms obtained prior to the pandemic. Given the unprecedented situation represented by the global pandemic and the hypothesised consequential impact on mental health relating to social distancing, lockdown measures and severe risk to health, it was felt that comparing data with pre-pandemic population norms could not be justified. Instead, two separate studies were conducted using the data obtained from the online questionnaire.

Comparing symptoms in children who do and do not stutter

In the first of these two studies, data collected from the stuttering sample was compared with a community cohort of children who do not stutter obtained over a similar period. Data for the non-stuttering group were obtained from a sub-sample of children who participated in the Surrey Communication & Language in Education Study (SCALES; Norbury et al., 2016). These children had completed the RCADS-25 over a similar time period, although the methods by which data were collected varied. The findings from this analysis highlighted some interesting differences between the two cohorts in terms of the effect of age and respondent on symptom scores. While data obtained from the stuttering group indicated that self-reported anxiety symptoms increased with age and that there was good agreement between self- and parent-reported anxiety and depression symptoms, the opposite was found for the non-stuttering group.

The decision to compare anxiety and depression symptoms in the stuttering cohort with a group of non-stuttering children who had completed the same mental health measure during the pandemic period, helped to control for the confound of the pandemic on population mental health. However, as this comparison had not been planned at the outset, it was not possible to control for some factors that may contribute to elevated internalising symptoms, namely co-occurring developmental conditions, and data collection practices.

Data from the stuttering group were obtained through an anonymous online questionnaire, while the non-stuttering group completed paper copies of the questionnaire in school or completed the RCADS-25 with the researcher in an online assessment session. Furthermore, group differences existed in mean age and socio-economic status, both of which are risk factors for adverse mental health. The limitations of collecting data during a pandemic should be recognised when interpreting these findings and scores in the stuttering group are not comparable with studies conducted prior to the pandemic. Nevertheless, it is worth noting that mean scores in both groups failed to reach clinical threshold, potentially hinting at the role of resilience in this age group in spite of unprecedented global events.

Examining factors associated with elevated anxiety and depression

The second study that utilised this questionnaire data examined the effect of several child (stuttering severity), family (family history of mental health or stuttering) and contextual (bullying victimisation) risk factors on anxiety and depression scores in the stuttering sample only. Family history of mental health problems significantly predicted anxiety and depression scores, with higher scores reported by those with a positive family history of adverse mental health. This would indicate a need to account for familial factors when examining the association between stuttering and internalising symptoms in young people in future research. Age was found to predict depression scores, with older children reporting higher scores, which is a trend also reported in the general population (Thapar, et al., 2012; Maughan & Collishaw, 2015). Bullying was associated with higher anxiety, but not depression, symptoms. However, bullying victimisation was only found to predict anxiety scores when age was removed from the model, which would suggest the effects of bullying may differ across age. The interaction of such child, family and contextual factors may change over the course of development, and therefore this would be an interesting question for future research.

As emphasised in chapters 3 and 4, the small sample sizes, presence of group differences in variables associated with mental health outcomes and

the confounding context of the pandemic limited the conclusions that could be drawn from this study. Nonetheless, these studies suggest that family history of mental health problems, older age and bullying victimisation are key indicators that increase risk for internalising difficulties in children and adolescents who stutter. This illustrates the importance of considering a broader range of familial factors when considering risk for elevated internalising symptoms in children who stutter, and the possibility that involvement of putative risk factors for adverse mental health may contribute towards the heterogeneity of symptom reporting in the extant literature.

Study 3: longitudinal secondary data analysis

Principally, the findings of the systematic review and meta-analysis as well as the empirical research highlighted the need for longitudinal, community studies that would examine internalising symptoms over the course of development in a non-clinical sample. The final study was designed in response to this observed gap in the literature. In order to mitigate the confounding impact of the pandemic, this study involved longitudinal analysis of secondary data obtained from the Millennium Cohort Study (MCS). This UK birth cohort study provided a sufficient sample size to permit examination of a broader number of factors that may be associated with internalising problems in a community sample. Creation of two grouping variables (stuttering/not stuttering; speech-language problems/no speech-language problems) facilitated examination of the effect of stuttering on internalising symptom scores, while accounting for co-occurring speech and language problems. Using parent-reported scores on the Strengths and Difficulties Questionnaire (SDQ) across six sweeps (age 3 – 17 years) of the MCS, it was possible to plot the symptom trajectories for four groups: children who stutter; children who had parent-reported speech and/or language problems; children who had a stutter and co-occurring speech and language problems; and children with no reported difficulties. Once confounding factors had been controlled for, there was no effect of stuttering on scores at time point one (age 3) nor in symptom trajectories over time. However, there was an effect of speech and language problems both in terms of obtaining, on average,

higher scores at age three and also a steeper trajectory over time. Cohort members who both stuttered and had speech and language problems presented with the highest internalising symptoms, indicating an additive effect of co-occurring speech and language difficulties in relation to stuttering.

Next, the risk for clinical-level internalising problems was examined by estimating group differences in report of deliberate self-harm and anxiety or depression diagnosis at age 17. The results from logistic regression models found no effect of stuttering on the odds of deliberate self-harm or clinical diagnosis of anxiety or depression at 17 years of age, suggesting that adolescents who stutter are no more likely to self-harm or develop clinical anxiety or depression than adolescents who do not stutter, after accounting for speech and language problems.

Finally, and in order to examine the cumulative effect of multiple factors on internalising problems in adolescents who stutter, five multiple risk models were reported. In all five models, female sex was associated with increased odds of internalising problems. It is worth noting that internalising scores were dichotomised in these analyses and therefore these findings relate to odds of scoring above the cut-off threshold rather than elevated internalising scores. There was some evidence that maternal mental health was also an important factor in clinical-level symptomology but the association was not as persistent across measures as female sex.

Given that the predictors included in the multiple risk models were all based on the existing literature pertaining to factors associated with risk for adverse mental health, it was surprising that so few predictors were found to be associated with increased odds of internalising problems. On the other hand, results of univariate analyses indicated a greater number of factors were associated with clinical-level scores, perhaps suggesting the importance of considering the amalgamated effect of a range of factors, and the interplay between them, when estimating risk for internalising problems.

In modelling multiple risk, it was hypothesised that this may enhance understanding of the heterogeneity in symptom reporting. Estimating the size of the effect of different predictors has provided insight into the extent to which certain factors may serve to increase risk for elevated symptoms beyond the average range. For example, many studies to date have included samples of children and adolescents of both sexes. The present findings would suggest that girls who stutter are more likely to present with elevated internalising symptom scores than boys.

Another consideration when examining risk for internalising problems and elevated symptomology in children and adolescents who stutter, is the mental health measure used. Studies comparing anxiety in children who do and do not stutter have relied on various mental health measures that may not all be assessing the same construct of anxiety. Different predictors may be associated with different mental health measures because of disparities in the underlying construct being assessed. Interestingly, being female was a persistent risk factor, whereas the extent to which maternal mental health was associated with risk for internalising problems was more variable across mental health measures. A further consideration is whether identification of risk factors varies across different respondents. For example, in this study a greater number of factors were associated with internalising problems on the parent-report measure in contrast with the self-report measures. The present research was not designed to consider respondent differences in symptom reporting, but this is an interesting question moving forward as it again may offer insight into the heterogeneity in symptom reporting.

Consequently, the third project attempted to address many of the methodological limitations identified in the systematic review of the existing literature and in the national online questionnaire studies. Still, limitations need to be considered when interpreting results of this final study. First, participants were assigned to groups on the basis of parent-reported difficulties. Therefore, the reliability of stuttering status is uncertain without clinical examination, meaning that some participants in this group may not have had a diagnosable stutter. Similarly, reliance on parent report for

assigning participants to the speech and language problems group is dependent on subjective judgement and knowledge of developmental milestones. Reported problems with speech and language also do not equate to meeting threshold for developmental delay or clinical disorder. Second, and particularly relevant to the stuttering group given the high recovery rate in early childhood, stuttering and speech-language status was dependent on parent-reported difficulties at a minimum of one sweep. While this permitted examination of group differences in internalising symptoms among individuals who had ever experienced difficulties with communication, it is likely that many individuals in both groups had resolved such difficulties by later sweeps. The decision to group participants in this way addressed issues around sample attrition in the MCS, which particularly in the case of the stuttering group, would likely have resulted in a substantially reduced sample size by the later sweeps, meaning that it would not be possible to conduct the proposed analyses due to insufficient statistical power. Third, as the MCS is not intended to study the stuttering population per se, unlike the ELVS Stuttering Study (Reilly et al., 2018) for instance, information about the nature of stuttering reported by an individual is sparse. For example, it was not possible to consider the influence of stuttering severity, time since onset, family history of stuttering and other stuttering-related factors on any association between stuttering and internalising problems. The wealth of child- and family-related data permitted inclusion of many putative risk factors for adverse mental health, which became the focus of the study.

Nevertheless, the findings from the multiple risk model contribute to our understanding of the size of the effect of particular predictors on the odds of internalising problems in this population. This provides a basis upon which to explore the interplay between a number of factors involved in risk for internalising problems in future studies.

Bringing it all together: are children and young people who stutter at risk of elevated anxiety and depression symptoms?

The association between anxiety and depression symptoms and developmental stuttering has been examined through three separate studies. A relatively consistent finding from these studies is the minimal evidence for any association between stuttering and clinical-level internalising problems among children and young people. On the other hand, the results of these three studies point to a mixed picture when it comes to risk for elevated internalising *symptoms* in this clinical population.

Children and adolescents who stutter do not differ in symptoms of depression, even after accounting for confounding factors. Comparison of symptom scores in children who do and do not stutter, before accounting for other factors, would suggest that children who stutter report higher anxiety symptoms than their peers. Once confounding factors are controlled for, there appears to be no effect of stuttering on anxiety symptom scores. While parent-reported scores were higher in the stuttering group after controlling for age and SES in one study (chapter 3), the model only adjusted for group differences in these demographic variables; there were a number of other putative risk factors that were not taken into account.

The present thesis has considered the role of a number of factors, both related to and independent of stuttering, which may go some way to explaining the heterogeneity in symptom reporting evidenced in the literature. In the following section those child, family and contextual factors that have been examined in the present research are discussed.

Factors associated with elevated anxiety and depression in childhood stuttering

It would seem reasonable that factors associated with increased risk for internalising problems in the general, non-stuttering population, would also be applicable to young people who stutter. This has been considered by Smith et al., (2014) who examined a number of putative risk factors

associated with anxiety in relation to childhood stuttering. They found little evidence that children and young people who stutter, and their families, were more likely to present with factors associated with increased risk for elevated anxiety in the general population. Results of the current systematic review found little evidence that these putative risk factors were accounted for when comparing mental health scores in children who do and do not stutter, even though they likely contribute to risk for internalising problems. While the current research cannot speak to environmental factors, apart from bullying, it has illustrated the importance of accounting for putative risk factors for adverse mental health in any evaluation of risk for greater internalising symptoms in young people who stutter. For instance, a child who stutters may obtain higher scores on a self-report measure of anxiety relative to a child who does not stutter, matched for socio-economic background and age. However, the child who stutters may have a family history of depression and experienced an adverse childhood event, whereas the non-stuttering child has not. In this event, it may be that the family history and adverse experiences, two acknowledged risk factors for adverse mental health, contribute to heightened emotional symptoms in this child over and above their stutter.

Child Factors

Co-occurring Speech, Language and Communication problems

Existing research has reported evidence of elevated anxiety among children and adolescents who stutter with co-existing speech, language and communication problems or other neurodevelopmental conditions (Blood et al., 2007; Briley, O'Brien & Ellis, 2019; Smith et al., 2017). Blood et al. (2007) and Briley et al. (2019) both compared adolescents who stutter with co-existing speech and language and non-speech and language problems, with children who stutter who did not exhibit co-existing difficulties. Both studies, one of which drew on a clinical sample and the other a community sample, found significantly higher anxiety symptoms reported in the group of adolescents who stutter with co-existing difficulties. Similarly, the findings from chapters two and five suggest that co-occurring communication

difficulties may be associated with risk for elevated anxiety symptoms in children and adolescents who stutter. The additive effect of co-occurring speech and language difficulties and stuttering in internalising symptom trajectories would also suggest that this group of children and adolescents need to be carefully monitored.

As discussed earlier in this thesis, elevated internalising symptoms have been observed in children with other neurodevelopmental conditions, including autism and ADHD (Gnanavel et al., 2019; White et al., 2009; Connor et al., 2003). The results from the MCS study hint at increased likelihood of elevated internalising problems in children who stutter who also have a diagnosis of autism as measured by parent-report. However, co-occurring autism diagnosis was not associated with elevated internalising problems on the other four measures of anxiety and depression symptoms, nor in terms of receiving a clinical diagnosis at 17 years.

Evidence to date would suggest co-occurring difficulties could be a moderating factor that should be taken into account in future studies investigating risk for anxiety in children and adolescents who stutter. Results presented in chapter five indicated that children who stutter who have co-occurring speech and language difficulties may be at particular risk for elevated internalising symptoms.

Many children who stutter show developmentally appropriate language skills (Watts et al., 2015; Watts et al., 2017; Nippold, 2018). Yet, co-occurrence of other speech, language and communication disorders in children who stutter has been well-documented, particularly amongst those individuals presenting for treatment (Arndt & Harley, 2001; Blood et al., 2003a; Briley & Ellis, 2018; Nippold, 2004; Unicom et al., 2020). Speech, language and communication difficulties encompass a broad variety of difficulties and conditions, and therefore further research is warranted to examine differences in the emotional and mental health profiles of children with distinct and co-existing communication problems.

The effect of age on symptom trajectories

Rates of anxiety and depression increase with age in the general population, with heightened risk in adolescence (Maughan & Collishaw, 2015). It has been suggested that many studies examining anxiety and depression symptoms in the stuttering population have encompassed broad age ranges in their samples, which may have contributed to inconsistent findings (Smith et al., 2014). As already noted, there are few studies involving pre-school children who stutter, which may be due to the difficulties in assessing anxiety in this age group and the focus on temperament. Most studies to date have involved school-aged children, and many of these studies have reported elevated anxiety, but not depression, symptoms in the stuttering group relative to the non-stuttering group. On the other hand, studies involving older adolescents and young people up to 25 years of age, have tended to report substantial differences between stuttering and non-stuttering groups (Doruk et al., 2008; Mulcahy et al., 2008), while adults who stutter appear to be at elevated risk for social anxiety disorder (Blumgart et al., 2010; Iverach & Rapee, 2014). Veerabhadrappe, Vanryckeghem & Maruthy (2021) concluded that speech-related anxiety is present in children who stutter by age seven and increases with age. However, the age at which children who stutter may be at particular risk for developing heightened internalising symptoms remains unclear.

The effect of age on risk for elevated anxiety or depression symptoms was examined in the present thesis. Although, the systemic review was unable to evaluate the influence of age on symptom reporting due to the small number of studies reporting anxiety symptoms across childhood, and lack of pre-school studies in particular, the effect of age was examined in the two other studies. Data obtained from the stuttering group in the online questionnaire study found that depression symptoms increased with age in this sample, although the age range was limited (8 - 15 years). The MCS study offered the greatest opportunity to examine symptom scores over time through examination of symptom trajectories. The symptom trajectories for all cohort members, regardless of whether they stutter or not, increased over the course of development.

It is likely that the extent to which age affects risk for internalising problems depends on the interaction with a number of other factors. For example, it may be that the relationship between bullying and internalising symptoms differs according to age, as hinted at in the analyses reported in chapter four.

The present research has not been able to enhance our understanding of the point in development at which children who stutter may be at elevated risk for internalising problems. Nevertheless, clinicians should be alert to the increased risk for internalising problems in mid- to late-adolescence when working with young people who stutter, especially girls.

Child sex

The mental health literature tells us that girls are more likely to report internalising problems than boys, particularly depression, and that this sex difference increases with age (Hankin et al., 1998; Kistner, 2009).

It is perhaps unsurprising then that analysis of the MCS data found female sex to be a consistent predictor of internalising problems in the stuttering population. Being female was associated with scoring above the clinical cut-off on all four self- and parent-report measures, as well as odds of being diagnosed with anxiety and depression by a health professional aged 17. Stuttering is much more common in boys than girls, with the sex ratio increasing with age. It is therefore important to recognise the increased risk that female sex poses for the mental wellbeing of young people who stutter. Monitoring anxiety and depression symptoms in female adolescents who stutter should be factored into clinical management.

Stuttering Severity

Synthesis of existing studies reported in chapter two suggested that stuttering severity may not be associated with anxiety symptom reporting. Empirical findings from the online questionnaire study also suggest that stuttering severity is not associated with increased risk for internalising

symptoms. This finding corroborates much of the earlier research which failed to find an association between stuttering severity and elevated anxiety. However, the way in which stuttering severity is measured may influence the extent to which it is associated with symptom reporting. It is likely that other environmental factors may play a role in the degree to which stuttering severity is associated with elevated internalising symptoms. For example, family support, strong friendships and supportive school environments may help to increase resilience and offset negative self-evaluation and internalising problems.

Family Factors

Socio-economic background

Lower socio-economic status has been associated with common mental health conditions, and these socio-economic inequalities in anxiety and depression appear to increase with age (Fryer et al., 2002; Green & Benzevel, 2013). As reported in chapter five, the effect of socio-economic status (SES) on internalising problems was assessed using three indicators of SES available in the Millennium Cohort Study: below OECD 60% median poverty indicator; maternal education (mother's NVQ-equivalent qualification); and OECD weighted income quintile. When focusing on the stuttering sample only, lower socio-economic status, as indicated by scoring below 60% median poverty indicator, was associated with elevated scores on the Kessler-6, SMFQ and parent-reported SDQ measures in univariate analyses, but these effects were not maintained once all variables were included in the model.

There are differences in the extent to which socio-economic indicators predict adolescent internalising mental health (Hazell et al., 2022). In this study, five SES indicators (parent education, household income, household wealth, parent occupational status, and relative neighbourhood deprivation) were associated with greater parent-reported internalising symptoms at age 14 and 17. In contrast, only three indicators (income, wealth and occupational status) were associated with self-reported internalising

symptoms at 14 and 17. Additionally, at both age 14 and 17, SES indicators predicted a greater amount of the variance in parent-reported symptom scores (4.73% and 4.06%, respectively) compared to self-reported internalising symptoms (0.58% and 0.60%, respectively). This led to the conclusion that the extent to which different socio-economic indicators predict adolescent mental health differs by respondent (Hazell et al., 2022). It may be that in the MCS study, the OECD poverty indicator variable selected for inclusion in the multiple risk factor models was not sufficiently sensitive to find an effect. Perhaps inclusion of another socio-economic indicator, such as household wealth, which Hazell et al. (2022) found explained the most variance in self-reported internalising scores, would have resulted in different findings.

Nevertheless, socio-economic status is likely to affect risk for internalising problems in the stuttering population, similarly to the non-stuttering population. As pointed out in the systematic review, many existing studies have examined anxiety in samples comprising middle-income families. This may relate to the reliance on clinically ascertained samples and the social gradient in access to services. Therefore, it will be important for future research to account for socio-economic differences in any investigation of internalising symptom reporting in stuttering and non-stuttering groups.

Parent mental health

Children of parents with anxiety or depression are at increased risk of developing internalising problems themselves (Rapee, 2015; Thapar et al., 2012). The association between family history of adverse mental health and elevated anxiety and depression symptoms was similarly observed in the present research. This is somewhat unsurprising given the extensive literature pertaining to the association between parental mental health and children's mental wellbeing. Nonetheless, as discussed in chapter two, family history of mental health problems have not routinely been reported in the field, suggesting these confounding variables have not been accounted for. The present findings serve to emphasise the need to control for family mental health when examining internalising symptoms in children who do

and do not stutter, and when assessing risk for internalising problems in clinical management.

Contextual Factors

Bullying

Bullying victimisation is associated with increased risk for poor mental health outcomes in childhood, and in the longer term. Given the documented experience of bullying in the stuttering population, it was hypothesised that being bullied would be associated with greater internalising symptoms. Previous research has pointed to an association between bullying, anxiety and stuttering (Blood & Blood, 2007; Cook & Howell, 2014). Findings from analysis of the online questionnaire data similarly illustrated an association between bullying victimisation and anxiety symptoms, although not depression symptoms. In the MCS analysis, participants were asked to disclose whether they had experienced bullying, and therefore differences in one's definition of bullying behaviour as well as response bias may have contributed to the null finding. When examining the effect of individual predictors on the odds of scoring above threshold on the SMFQ and Kessler-6, bullying almost reached statistical significance ($p = .05$).

The body of literature showing the detrimental impact of childhood bullying on mental health and wellbeing in the non-stuttering population (Moore et al., 2017; Reijntjes, et al., 2010) combined with the reportedly high rates of bullying amongst adolescents who stutter (Blood & Blood, 2004; Blood & Blood, 2007; Erickson & Block, 2013; Langevin et al., 1998), would suggest a need for schools and health services to pursue anti-bullying initiatives.

In summary, a number of factors are associated with risk for internalising problems in adolescence, including family history of mental health problems, childhood adverse experiences, bullying victimisation, sex (internalising problems are higher in girls), low socio-economic status, and peer relationship difficulties (Fryer et al., 2002; Green & Benzevel, 2013; Narmandakh et al., 2020; Pine & Klein, 2015; Rapee, 2015; Thapar et al.,

2012). It is therefore perhaps unsurprising that many of these factors were also found to be significantly associated with internalising symptoms in the present research. However, as highlighted by the systematic review, often this information is either not collected or not controlled for in studies examining the association between stuttering and anxiety/depression. Consequently, it may not be stuttering per se which is associated with elevated risk for internalising problems, but differences in, and the interplay between, a number of other familial, child and environmental factors.

Clinical implications

The present findings emphasise the need for clinicians to be alert to the range of factors that may put children who stutter at risk for poor mental health outcomes and to ensure that promotion of mental health and well-being is embedded into the management of children who stutter from an early age.

Thorough case histories that gather information about parental mental health and family factors, which may suggest a child is at increased risk for poorer mental health outcomes, would help in planning appropriate and holistic care plans. While resources are often stretched in clinical settings, completing short mental health screens with parents may be one way to identify potential risk related to familial factors but also the ways in which clinicians can facilitate parents in supporting their child over the course of intervention. In their consensus guidelines, Brundage et al. (2021) recommend six core assessment areas when evaluating stuttering. Speech, language and temperament development, and the adverse impact of stuttering on quality of life, employment and education are considered core areas for assessment. Yet, based on the present findings, assessment of internalising symptoms and obtaining information about family factors, such as family history of mental health issues, would also be valuable components of comprehensive assessment. There are a range of screening tools for child and adolescent mental health, which are routinely used in clinical and research settings (see Fisher, Chin & Vidair, 2015), although it is acknowledged that tools more

closely aligned to the psychological impact of stuttering on school-age children are lacking (Jones et al., 2021).

Increased awareness of the risk for bullying and the impact of stuttering on peer relationships and social development, among education practitioners, would help to create a more inclusive environment in which those context-related risks can be mitigated. Similarly, awareness of the likely impact of increasing age on internalising symptoms is an important consideration when working with adolescents who stutter, both in terms of the transition to secondary school as well as the pressures associated with transitioning into early adulthood. Embedding resilience, self-esteem and confidence-building into the management plan for adolescents who stutter may help to off-set internalising problems that interfere with social functioning. Many researchers have advocated for a comprehensive approach to stuttering therapy for school-aged children and adolescents, which focuses on fluency, cognitive-behavioural elements and other goals that address the broader experiences of stuttering to mitigate the impact of stuttering on psychosocial and quality of life outcomes (Craig, 2003; Yaruss et al., 2008). Although several therapy approaches for adolescents incorporate cognitive elements, including emotional well-being and communication attitudes, currently there is insufficient robust evidence supporting the effectiveness of interventions for adolescents who stutter (see Baxter et al., 2016; Brignell et al., 2021).

Finally, being alert to the possibility of emotional problems in adolescence has the potential to increase early identification and intervention to mitigate the longer-term, deep-seated effects of poor mental health. Multi-disciplinary support for children and young people who stutter could facilitate more effective, targeted support for their specific needs. Greater collaboration between speech and language therapists, psychologists and teachers would ensure that young people who stutter benefit from the specialist support offered by respective professions. Furthermore, multi-disciplinary collaboration would help to embed the support mechanisms the child finds most beneficial into the different contexts in which they spend much of their time. Combining different professional perspectives and approaches could

facilitate delivery of targeted intervention programmes and opportunities to generalise coping strategies to develop self-confidence, resilience and self-esteem.

Theoretical Implications

Over the decades, researchers have suggested that anxiety causes stuttering (Sheehan, 1970); anxiety is a consequence of stuttering (Perkins, 1979; Ryan, 1974); and that anxiety may play a mediating role in the development of stuttering (Brutten & Shoemaker, 1967).

The research reported here does not support the hypothesis that anxiety causes stuttering. The variability in symptom reporting, both across the published literature, and within the present studies serves to emphasise that not all children who stutter experience elevated symptoms of anxiety and/or depression. Additionally, despite the limited number of pre-school studies, to date there is little evidence of elevated anxiety or depression symptoms in early childhood stuttering (Park et al., 2021; van der Merwe et al., 2011), which would suggest that anxiety is not present at stuttering onset. Furthermore, the results from the MCS study are consistent with the findings from a limited number of community studies in the field, showing that children and adolescents who stutter do not differ in anxiety symptoms. Should anxiety be causally related to stuttering, one would not expect such heterogeneity in symptom reporting during the course of development.

The DSM-5 (APA, 2013) definition for Childhood-Onset Fluency Disorder (stuttering) states that stuttering “causes anxiety about speaking...” (p46). The current findings would suggest that the relationship is more complex, and involves an interplay between a number of child-, family- and contextual-factors. To begin with, not all children who stutter go on to report elevated anxiety symptoms, as evidenced in the present thesis. In fact, examination of internalising symptom trajectories in chapter five does not support the hypothesis that children who stutter are any more likely to develop elevated internalising symptoms over and above non-stuttering peers, after

accounting for confounding factors. Instead, co-occurrence of stuttering and speech and language problems are more likely to be associated with elevated internalising symptoms in early childhood and throughout development. Finally, the present findings indicate that once factors associated with increased risk for internalising problems have been taken into account, there is little evidence of an effect of stuttering on anxiety symptom scores.

Instead of a causal relationship, it would appear that stuttering and anxiety may co-occur depending on the unique interplay of factors specific to that individual. Across the three studies presented, there is evidence that female sex and family mental health problems in particular, as well as bullying victimisation, co-occurring speech and language problems, and older age, are important predictors in risk for elevated internalising symptoms in children and adolescents who stutter. The extent to which adolescents who stutter experience stigma and negative reactions to their speech will vary (see Blood et al., 2003b; Erickson & Block, 2013; Boyle, 2018), and the degree to which these experiences have an impact on one's mental health will depend on the interaction between risk and protective factors (Rutter, 2009). This PhD research did not focus on many of these environmental factors, apart from bullying, and therefore how they interact with other child- and family- factors is an area for future research.

Implications for public health policy

In light of the present findings, public health messaging should focus on the importance of earlier identification and onward referral of children who stutter to mitigate the risk for development of internalising symptoms in this population.

Over the past few decades, there has been a shift away from 'wait and see' approaches to early childhood stuttering, and it is increasingly recognised that accessing specialist support near onset can be beneficial both to the child and the family as a whole (Snijders, et al., 2023; Kefalianos et al.,

2022). Families often turn to their GP or educational setting for advice and support when they have concerns about their child's development. Since primary care providers and schools typically function as referrers to speech and language therapy services, it is important they have sufficient knowledge and understanding of the condition and the support available. For instance, universal delivery of information to primary care services would empower GPs to make earlier referrals to appropriate tertiary services. Similarly, incorporating information about the impact of childhood stuttering into the training programmes of early education professionals, teachers and teaching assistants would increase confidence in seeking appropriate professional support.

It is clear that children who stutter are a heterogeneous group and therefore their experiences of both the overt characteristics of stuttering and the extent to which stuttering impacts their social and emotional development will vary. An awareness raising campaign that aims to promote greater understanding of the condition and the potential adverse impact on children's mental health and wellbeing, would facilitate earlier identification, timely and appropriate referral.

Limitations of the thesis

The research presented in the present thesis aimed to further examine, and extend our understanding of, the association between childhood stuttering and risk for anxiety and depression during development. Despite efforts to mitigate the confounds of the pandemic context, it must be acknowledged that the evidence from a cohort of children who stutter collected during this period cannot be compared to pre-pandemic studies.

Reflecting on the online questionnaire study, there are several changes I would make to the protocol. In chapter three, I provide rationale for comparing the stuttering group with a group of non-stuttering children involved in the SCALES study. The changes made to the pre-registration for this chapter were made in an effort to compensate for the confounds of the

pandemic. However, comparing RCADS-25 scores with population norms may have precluded some of the challenges encountered with interpretation of the findings reported in chapter three. Differences in administration and timing of data collection between the SCALES study and the online questionnaire introduced confounding variables, such as group differences in age, SES, existence of co-occurring developmental problems, and geographical location of the samples. While efforts were made to counteract anticipated group differences by matching participants, this was hampered by the requirement for full datasets from a limited pool of children assessed during a similar timeframe in the SCALES study. Under different circumstances, it would have been preferable to either recruit a second sample of typically developing children who do not stutter to complete the same online questionnaire within the same timeframe, or to compare the stuttering group with population norms.

With hindsight, I would have included a measure of the impact of stuttering on the child, for instance the Overall Assessment of the Speaker's Experience of Stuttering (OASES), alongside a clinical measure of stuttering frequency, such as the Stuttering Severity Instrument, in place of a non-standardised self-reported severity scale. This would have permitted closer examination of some of the questions that were discussed in chapter four pertaining to whether the way that stuttering severity is measured affects any association with anxiety and/or depression symptom scores. For example, I could have compared the association between RCADS-25 scores and scores on a standardised self-report versus clinician-report of stuttering.

In addition, I would have selected a bullying measure that was more sensitive to the experiences of childhood stuttering. For instance, the Teasing/Bullying Questionnaire for children who stutter (Langevin et al., 1998) would have enhanced understanding of the extent to which children in this sample faced bullying related to their stutter. Bullying is associated with adverse mental health in non-stuttering children (Lereya et al., 2015; Schoeler et al., 2018). Therefore, understanding how stuttering-related teasing and bullying affects mental health and well-being would have more

direct clinical and policy implications; informing practical interventions aimed at tackling negative peer reactions in children who stutter specifically.

While the purpose of this doctoral research was never to estimate rates of anxiety and/or depression disorder in children who stutter, the inclusion of more than one mental health measure would have been beneficial. One of the limitations cited in the systematic review is the fact that studies to date have employed a variety of measures that likely tap different anxiety constructs. While the RCADS-25 allowed me to collect data on anxiety and depression symptoms within one measure, and from both child- and parent-respondents, another measure of internalising symptoms would have been helpful in estimating risk. As shown in chapter 5, the factors associated with above-threshold symptom scores differed across mental health measure. Supplementing the RCADS-25 with another measure of anxiety and depression would have offered an opportunity to compare symptom reporting across measures.

Information on co-existing speech, language and communication difficulties was not collected in the online questionnaire. This is a question that would have permitted examination of the relationship between co-occurring communication difficulties and stuttering, which is a central question in my thesis. This information was also available in the SCALES cohort, and therefore should those data have been available for the stuttering group and had the sample been larger, it may have been possible to perform sub-group analyses.

Finally, reliance on secondary data from the MCS meant that I had no influence over the variables collected from a population cohort, and therefore this limited the questions I could reasonably address. Ideally, in order to more robustly examine the risk for internalising symptoms in children who stutter across the course of development, I would have employed a longitudinal study design, following children from the point at which they begin to stutter through to early adulthood. This would have been outside the scope of a four-year doctoral programme. Nevertheless, resources and time

permitting, a longitudinal population study would have permitted me to select measures that assessed covert and overt characteristics of stuttering, in addition to family-, child- and environmental-factors, facilitating a more valid and precise estimate of risk for internalising symptoms in the community.

Directions for future research

(i) Risk mechanisms

The present research has shown there to be an effect of particular factors on elevated internalising symptoms in children and adolescents who stutter, notably female sex, parent mental health, co-occurring speech and language problems, and age. These findings will require replication and expansion in future studies. There are many other factors that could not be explored in this research, which could potentially influence risk for internalising problems. Furthermore, the extent to which particular factors in the context of other risk and protective factors, contribute to likelihood of developing internalising problems would facilitate more effective clinical management. This includes differentiating distal and proximal risk factors; factors associated with increased risk for psychopathology may be directly or indirectly affecting psychopathological risk (Rutter, 2009). It is also important to consider the mediating mechanisms that may underly causal effects, rather than focusing purely on identification of risk factors associated with psychopathology (Rutter, 2009).

(ii) Protective Factors & Resilience

Regrettably, a topic that has received comparatively little focus in my thesis is the role of protective factors and resilience in the development of internalising problems in this population. This remains a question for future research, especially given the heterogeneity of symptom reporting across studies. Variation in mental health outcomes can be attributed to the effects of both risk and protective factors. Conversely, resilience refers to positive psychological outcomes in the face of serious adversity and is concerned with the variation among individuals in response to the same level and type of adversity (Rutter, 2006; Rutter, 2009).

In the present thesis, risk factors have primarily been the focus of examination. Chapters two and four touch on the importance of protective as well as risk factors when examining internalising problems in children who stutter, although the hypothesis that positive family history of stuttering would act as a protective factor was not supported in the online questionnaire data. Researchers in the field of psychology have re-focused efforts when investigating risk mechanisms, by considering both the nature of risk exposure and the coping mechanisms employed in this context (Rutter, 2009). Consequently, there is scope for future research to consider factors that may protect against poor mental health and the interplay between risk and protective factors. This would also be beneficial when thinking about facilitating resilience in children who stutter to help them manage stress and adversity, which may stem from social situations.

In the general population, many children considered at high risk of depression and anxiety, for example due to genetic predisposition or childhood adversity, do not go on to experience adverse mental health (Rutter, 2009). The substantial individual differences in the way that one responds to stress and adversity have partly been attributed to the interaction between genetic and environmental factors, and also the role of resilience (Rutter, 2009).

Resilience depends on the interaction of a number of genetic, environmental and biological factors, which occur at the individual, family and community level (see Fritz et al., 2018; Huberty, 2012; Ungar & Theron, 2020). A recent systematic review examined studies reporting on the emotional, social, cognitive and behavioural resilience factors that may attenuate psychopathology in adolescents who have experienced childhood adversity (Fritz et al., 2018). They found evidence for 13 individual-level resilience factors (including cognitive, emotion regulation, social interaction/attachment and personality/self-concept factors); six family-level resilience factors (including family support and parenting factors); and one community-level factor (high social support). Consequently, resilience in mental health can be

considered in a systemic framework given the interaction of a variety of factors at the individual-, family- and community-level (Fritz et al., 2018; Ungar & Theron, 2020).

In terms of anxiety and depression specifically, research has identified a number of protective factors that may be associated with resilience.

Resilience in development of depression may be associated with good quality social relationships, effective coping mechanisms, emotional regulation, inherited factors and high intelligence (Thapar et al., 2012).

Positive family environments, self-efficacy and social support were identified as protective factors associated with fewer depressive symptoms in childhood in the BELLA study (Klasen et al., 2015). Social support and coping skills have been identified as protective mechanisms in development of anxiety (Donovan & Spence, 2000).

Protective factors in stuttering

The influence of protective factors in off-setting adverse mental health outcomes has been the focus of several studies looking at adults who stutter. Craig, Blumgart and Tran (2011) isolated several factors that could potentially protect adults who stutter from adverse mental health, including social support, healthy social functioning and self-efficacy. Blumgart, Tran and Craig (2014) found that adults who stutter who had poorer social support had elevated depressive mood and anxiety compared to adults who stutter with a high amount of social support. Craig, Blumgart and Tran (2015) reported that self-efficacy appears to play a protective role in negative mood states in adults who stutter, contributing to the variability in mood states in this population. Social support accounted for a comparatively smaller amount of the variance in mood state over time, however stuttering variables, such as percentage syllable stuttered (%SS), were not associated with mood state (Craig et al., 2015).

Interestingly, a recent study found a significant association between resilience and covert, but not overt, characteristics of stuttering (Freud &

Amir, 2020). This was evidenced by the fact that subjective experiences of stuttering, as measured on the OASES, were negatively correlated with resilience scores, while stuttering severity, measured using the Stuttering Severity Instrument (SSI), was not associated with resilience scores in a group of adults who stutter ($n = 30$). Grobbel (2022) reported that greater resilience predicted less adverse impact of stuttering on school-aged children and adolescents.

As these studies highlight, equally important in any discussion pertaining to risk for adverse mental health are the issues of protective factors and resilience. Improving our understanding of which factors serve to offset risk for internalising problems and enhance resilience, could help guide psychosocial management of children who stutter and arm them with the skills to become resilient adults. However, many published studies examining anxiety in children who stutter either have not reported or have not controlled for the effect of these potential moderating factors, which would inform clinical understanding of the underlying risk for development of anxiety and/or depression in stuttering.

Although direct examination of hypothetical protective factors has not been feasible in the current thesis, a number of factors have been implicated in the association between stuttering, anxiety and depression in children. Findings across the three projects discussed serve to emphasise the heterogeneity of children who stutter, especially in terms of their experiences of anxiety and depression symptoms. Such variability in psychosocial outcomes likely reflects the interaction of several factors – some of which are known to be associated with emotional disorders in the general population, and some of which are related to the negative social and environmental experiences associated with stuttering. While much of the current thesis has focused on factors that may serve to increase risk, further research is required to explore the contribution of protective factors that promote resilience in children who stutter. This could enhance our understanding of the complex interplay between variables that contribute to the association between stuttering and emotional wellbeing. Furthermore, such knowledge would provide

opportunities in clinical practice to identify strengths to promote the mental well-being of children who stutter close to onset. While resilience cannot be explicitly taught, there are ways in which young people can be supported to prepare themselves for dealing with adversity and thus make resilience more likely (Rutter, 2015).

(iii) Stuttering Recovery and Persistence

Another question for future research is whether there are differences in internalising symptom reporting among children and young people who recover from stuttering and those who persist into adulthood. In the adult literature, rates of social anxiety disorder are estimated around 40% (Blumgart, Tran & Craig, 2010). However, a question for future research is whether ever experiencing stuttering during childhood increases risk for elevated internalising symptom, even after the stutter has resolved. The MCS study attempted to address this question, but findings require replication in community samples in which stuttering can be more accurately diagnosed and recovery closely monitored.

(iv) Stuttering intervention and mental well-being

The findings reported in this thesis indicate that some children and adolescents who stutter will develop elevated anxiety and/or depression symptoms. This could have implications for treatment, both in terms of efficacy and maintenance of therapeutic outcomes. The broader impact of adverse mental health on social functioning, educational and physical health outcomes (Jamnik & DiLalla, 2019; von Simson et al., 2021; Liu, Chen & Lewis, 2011) may create barriers to achieving therapeutic objectives and generalisation to non-clinical contexts. For instance, children who stutter who experience elevated anxiety may be less likely to enter unfamiliar social situations, and therefore do not have the opportunity to practice speech fluency techniques or build confidence in speaking with other children.

In a study involving adults who stutter ($n = 64$), Iverach et al. (2009b) found that presence of mental health disorders impacted stuttering frequency and

situation avoidance outcomes following speech-restructuring treatment. Existence of any mental health disorder was associated with poorer outcomes in the short term and 6-months post-therapy (Iverach et al., 2009b). This study highlights the impact that mental health disorders may have on the anticipated outcomes of speech and language therapy, as well as the extent to which clients are able to maintain these outcomes in the longer-term. While the research findings in this thesis do not lend support to greater likelihood of clinical-level symptomology in school-aged children and adolescents who stutter, diagnostic clinical assessments for mental health conditions were not employed to confirm presence or absence of anxiety and mood disorders. Nonetheless, elevated anxiety and depression symptoms may still affect achievement and maintenance of therapy outcomes, even if these symptoms are sub-clinical.

The potential impact of concomitant mental health difficulties in childhood stuttering on effectiveness of intervention also underscores the importance of thorough assessment prior to commencement of stuttering intervention. For example, it may be beneficial to address mental health issues before beginning speech and language therapy in order to optimise maintenance of therapeutic benefits for stuttering (Iverach et al., 2009b). Future research that evaluates the extent to which sub-clinical anxiety and depression symptoms are associated with immediate and long-term therapy outcomes would enhance clinical management of stuttering.

Poor mental health in children who stutter may also have implications for effective implementation of family-focused intervention approaches. A child's emotional and behavioural problems can negatively impact family life, for instance effecting family routines, parents' quality of life and well-being, and the relationship between parents and extended family members (Early, Gregoire & McDonald, 2000; Crowley & Kazdin, 1998; Farmer et al., 1997). Consequently, poor mental health in children and the associated impact on family functioning and parent well-being may affect the extent to which parents can engage in and implement family-oriented interventions. Therapy approaches for pre-school stuttering typically rely on high levels of parental

involvement, for instance designated time on parent-child interaction strategies, daily practice of fluency techniques, and adaptations to the child's home environment to induce greater fluency (see Baxter et al., 2016; Brignell et al., 2021; Laiho et al., 2022). Parents may find it difficult to implement such strategies with their children in the context of adverse child mental health and disrupted family functioning. Research examining not only the impact of *stuttering* on the wider family, but also the impact of *concomitant internalising problems* alongside stuttering on the family and therapy efficacy could enhance our understanding of effective management and the mechanisms that may be involved in stuttering relapse.

Finally, the degree to which therapy approaches for childhood stuttering facilitate positive change in emotional well-being, and the mechanisms underlying treatment effects, are important considerations for future research. The research presented here would suggest that stuttering severity is not associated with internalising symptom reporting. This may mean that intervention approaches aimed purely at reducing or eliminating moments of stuttering will not facilitate positive changes in the mental health and wellbeing of children who stutter. On the other hand, it may be that achieving greater levels of fluency could lead to improved confidence around speaking in social contexts, resulting in increased self-esteem and more positive self-evaluation.

Evidence comparing a direct (Lidcombe Program) and indirect (Rotterdam Evaluation Study of Stuttering Therapy; RESTART-DCM) therapy approach for pre-school children who stutter, found similar outcomes in terms of fluency, health-related quality of life, children's speech attitudes and emotional and behavioural symptoms in the two treatment groups (de Sonnevile-Koedoot, et al., 2015). Interestingly, the Lidcombe Program aims to decrease stuttering frequency based on operant conditioning principles, whereas RESTART-DCM involves modification of the child's environment to induce fluency, suggesting that direct work on fluency may facilitate improvements in emotional well-being. Yet, a recent Cochrane Review of pre-school interventions for stuttering was unable to report on outcomes

pertaining to emotional, social and psychological domains or communication attitudes associated with the extensively evidenced Lidcombe Program (see Sjøstrand et al., 2021). Hancock et al. (1998) reported on the long-term benefits of three different fluency-shaping therapy approaches on stuttering frequency, anxiety and communication attitudes in school-aged children and adolescents who stutter.

These findings may suggest an indirect impact of fluency-shaping approaches on the emotional wellbeing of children. However, insufficient evidence is currently available for school-aged and adolescent intervention approaches for stuttering, and more robust research is warranted for many of the pre-school interventions currently available which do report on social and emotional outcomes (see Laiho et al., 2022; Brignell et al., 2021; Sjøstrand et al., 2021). Nonetheless, measures of mental health and wellbeing are important additions to the bank of outcome measures selected by speech and language therapists working with children and adolescents who stutter, and future research into intervention efficacy.

(v) Qualitative approaches

The present thesis utilised quantitative approaches to address the research questions set out in chapter one. However, there is also a place for qualitative methodologies to expand on the data presented here. For instance, gathering qualitative data to understand the experiences of children and young people who stutter in school could provide more detailed understanding of factors that affect social and emotional development. These experiences could inform development of resources for schools to ensure children receive effective support in educational settings. Gathering data about families' experience of accessing specialist services and perspectives on the availability of support would help to paint a picture of the strengths and areas for improvement in public health provision. Symptom scales and questionnaires could be complemented by qualitative approaches that enrich our understanding of the challenges young people and their families face, informing policy recommendations and clinical guidance.

Conclusion

The present thesis asked whether children and adolescents who stutter are at risk for increased anxiety and depression symptoms relative to peers who do not stutter. This research found limited evidence that stuttering is associated with elevated symptoms of either anxiety or depression in the community context, although children who access clinical services may be more likely to present with internalising problems (Iverach et al., 2016).

The overarching finding from this research is the fact that children who stutter are not a homogenous group; individual and family differences need to be acknowledged in terms of both the experience and impact of stuttering on child and adolescent mental health and emotional development. Through examination of the existing literature and analysis of community data it would appear a number of factors serve to increase risk for elevated internalising symptoms in the stuttering population. The complex interplay between child, family and contextual factors requires investigation in longitudinal, population studies. Nevertheless, clinicians should be particularly alert to co-occurring speech and language problems, family history of adverse mental health, female sex and bullying when evaluating risk for poorer emotional outcomes. Holistic assessment will help to guide preventative, rather than reactive, management to offset internalising problems that have the potential to hinder lifelong potential.

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Appendices

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 (see below).

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

Initial Rapid Screen Form

1. Is this reference relevant for our review? (At first 'no' answer, select submit)

- Yes
- No; not published in English
- No; not developmental stammering
- No; participants outside age range (<2;0 OR >25;0)
- No; no control group of non-stammering participants
- No; study design other than RCT, QED, longitudinal, cohort study, cross-sectional (e.g. review, theoretical paper, case study)
- No; study does not refer to conditions DSM-5 classified under 'anxiety disorders' or 'depression disorders'
- Can't tell (comment optional)

Full Text Screening Form

1. Should this study be included in the review?

- Yes
- No; not published in English
- No; not developmental stammering
- No; participants outside age range (<2;0 OR >25;0)
- No; does not refer to conditions DSM-5 classifies as 'anxiety disorders' or 'depressive disorders'
- No; does not include non-stammering comparison group
- No; study design is not an RCT, quasi-experimental design; longitudinal, cohort or cross-sectional study
- No, other

Data Extraction Form

RefID: 1, There and Back Again: A Review of Residency and Return Migrations in Sharks, with Implications for Population Structure and Management. Chapman DD, Feldheim KA, Papastamatiou Y, Hueter RE

The overexploitation of sharks has become a global environmental issue in need of a comprehensive and multifaceted management response. Tracking studies are beginning to elucidate how shark movements shape the internal dynamics and structure of populations, which determine the most appropriate scale of these management efforts.

Tracked sharks frequently either remain in a restricted geographic area for an extended period of time (residency) or return to a previously resided-in area after making long-distance movements (site fidelity). Genetic studies have shown that some individuals of certain species preferentially return to their exact birthplaces (natal philopatry) or birth regions (regional philopatry) for either parturition or mating, even though they make long-distance movements that would allow them to breed elsewhere. More than 80 peer-reviewed articles, constituting the majority of published shark tracking and population genetic studies, provide evidence of at least one of these behaviors in a combined 31 shark species from six of the eight extant orders.

Residency, site fidelity, and philopatry can alone or in combination structure many coastal shark populations on finer geographic scales than expected based on their potential for dispersal. This information should therefore be used to scale and inform assessment, management, and conservation activities intended to restore depleted shark populations. Expected final online publication date for the Annual Review of Marine Science Volume 7 is January 03, 2015.

Primary Outcomes

1. Study design

- ☐ Randomised control trial (RCT)
- ☐ Quasi-experimental design
- ☐ Longitudinal design
- ☐ Cohort study
- ☐ Cross-sectional study (with control group)
- ☐ Other appropriate design (with control group)

Clear Response ()

2. Mental health condition(s) the study is focusing on

3. Assessment measure(s) used to assess anxiety / depression

4. Method of identifying stammer

5. State the definition of stammering used by the author

6. State the definition of anxiety used by the author

7. State the definition of depression used by the author

8. Number of participants

- ☐ Stammering group
- ☐ Non-stammering control group
- ☐ Total number of participants

9. Measure used to assess severity of stammering (tick all that apply)

- ☐ % syllable stuttered
- ☐ severity rating (severe, moderate, mild, etc.)
- ☐ Likert type scale (e.g. 1 - 10)
- ☐ Published assessment measure
- ☐ Comment

10. How is reported socio-economic status measured?

Postcode/area

- ☐ Proportion of free school meals
- ☐ Household income
- ☐ Parental job status
- ☐ Other (please comment)
- ☐ Not reported

11. Overall socio-economic status for sample

(e.g. 40% free school meals; mean status)

- ☐ Reported socio-economic status of all participants
- ☐ Reported socio-economic status for stammering group (if available separately)
- ☐ Reported socio-economic status for non-stammering control group (if available separately)
- ☐ Not reported

12. Sex of participants (stammering group)

Female

Male

Not reported

13. Sex of participants (non-stammering group)

- ☐ Female
- ☐ Male
- ☐ Not reported

14. Age groups of participants

- ☐ Stammering group age (mean / range)
- ☐ Non-stammering group age (mean / range)

15. Presence of co-morbid disorders (stammering group)

- ☐ Yes (number with co-occurring disorders)
- ☐ No (number without co-occurring disorders)
- ☐ Not reported

16. Presence of co-morbid disorders (non-stammering group)

- ☐ Yes (number with co-occurring disorders)
- ☐ No (number without co-occurring disorders)
- ☐ Not reported

17. First language of participants

- ☐ Stammering group
- ☐ Non-stammering group
- ☐ Not reported

18. Comment on languages represented in study sample

19. There is a family history of stammering (stammering group only)

- ☐ Yes (no. of participants)
- ☐ No (no. of participants)
- Not reported

20. There is a family history of mental health conditions (stammering group only)

- ☐ Yes (no. of participants)
- ☐ No (no. of participants)
- ☐ Not reported

21. There is a family history of mental health conditions (non-stammering group only)

- ☐ Yes (no. of participants)
- ☐ No (no. of participants)
- ☐ Not reported

22. Measures of anxiety (stammering group) - include all measures relevant

- ☐ Name of measure
- ☐ Mean
- ☐ Standard Deviation
- ☐ Not reported (comment)
- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation
- ☐ Not reported (comment)
- ☐ If further measures to be included, detail here (M, SD)

23. Measures of anxiety (non-stammering group) - include all measures relevant

- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation
- ☐ Not reported (comment)
- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation
- ☐ Not reported (comment)
- ☐ If further measures to be included, detail here (M, SD)

24. Measures of depression (stammering group) - include all measures relevant

- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation
- ☐ Not reported (comment)
- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation
- ☐ Not reported (comment)
- ☐ If further measure included detail here (M, SD)

25. Measures of depression (non-stammering group) - include all measures relevant

- ☐ Name of measure
- ☐ Mean
- ☐ Standard deviation

☐ Not reported	
☐ Name of measure	
☐ Mean	
☐ Standard deviation	
☐ Not reported	
☐ If further measures to be included, detail here (M, SD)	

26. Group comparisons on mental health measures

☐ Measure #1 (name)	
☐ P-value	
☐ t-test value (if applicable)	
☐ F-value (ANOVA)	
☐ Type of Effect Size calculated	
Effect Size difference between stammering & control groups	
Measure #2 (name)	
P-value	
☐ t-test value (if applicable)	
☐ F-value (ANOVA)	
☐ Type of Effect Size calculated	
☐ Effect Size difference between stammering & control groups	
☐ Measure #3 (name)	
☐ P-value	
☐ t-test value (if applicable)	
☐ F-value (ANOVA)	
☐ Type of Effect Size calculated	
☐ Size of Effect Size difference between stammering & control groups	
☐ Measure #4 (name)	
☐ P-value	
☐ t-test value (if applicable)	
☐ F-value (ANOVA)	
☐ Types of Effect Size calculated	
☐ Effect size difference between stammering & control groups	

27. The article is useful for:

☐ Qualitative analysis

☐ Quantitative analysis

☐ Comment

Appendix B: Risk of Bias

Table 21: 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/>
Post-publication addition in response to thesis examiners:

The Busse & Guyatt tool used to assess risk of bias is based on the [Newcastle-Ottawa Scale](#) and the Cochrane risk of bias instrument. Questions were adapted to ensure relevance to included studies <https://www.distillersr.com/resources/methodological-resources/risk-of-bias-in-cohort-studies>

Appendix C: Table of excluded studies

Table 22: Example of reasons for excluding studies

Study	Reason for exclusion
Blood, G. W., Blood, I. M., Tellis, G., and Gabel, R., 2001. Communication apprehension and self-perceived communication competence in adolescents who stutter. <i>Journal of Fluency Disorders</i> , 25, 161-178.	No evidence-based measure of anxiety
Erickson, S., and Block, S., 2013. The social and communication impact of stuttering on adolescents and their families. <i>Journal of Fluency Disorders</i> , 38, 311 – 24.	No control group; no evidence-based measure of anxiety
Gunn, A., Menzies, R. G., O'Brian, S., Onslow, M., Packman, A., Lowe, R., Iverach, L., Heard, R., and Block, S., 2014. Axis I anxiety and mental health disorders among stuttering adolescents. <i>Journal of Fluency Disorders</i> , 40, 58 – 68.	No control group
Kefalianos, E., Onslow, M., Ukoumunne, O., Block, S., and Reilly, S. 2014. Stuttering, temperament, and anxiety: Data From a community cohort ages 2-4 years. <i>Journal of Speech, Language, and Hearing Research</i> , 57, 1314-1322.	No evidence-based measure of anxiety (temperament)
McAllister, J., 2016. Behavioural, emotional and social development of children who stutter. <i>Journal of Fluency Disorders</i> , 50, 23-32.	No evidence-based measure of anxiety (temperament)
Smith, K. A., Iverach, L., O'Brian, S., Mensah, F., Kefalianos, E., Hearne, A., and Reilly, S., 2017. Anxiety in 11 year old children who stutter: findings from a prospective longitudinal community sample. <i>Journal of Speech, Language, and Hearing Research</i> , 60, 1211-1222.	No evidence-based measure of anxiety; lack of non-stuttering control group for the measure of anxiety (Spence Children's Anxiety Scale)

Appendix D: Studies excluded from meta-analysis

Details of studies excluded from the meta-analysis but which met criteria for the systematic review can be seen in the table below. Andrews and Harris (1964) reported the proportion of participants receiving a given score on the General Anxiety Scale for Children (GASC; Sarason et al., 1960) rather than group means, which meant the study could not be included in the meta-analysis. Interestingly, Andrews and Harris (1964), the only community sample of these three studies, reported no significance differences in reported anxiety between groups. However, scores on the Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2001) were found to be significantly higher in the stuttering group in both the Giorgetti et al. (2015) and Tiğrak et al. (2020) studies. As it is unclear whether group differences reflect elevated anxiety or depression symptoms, these group differences are more difficult to interpret.

Table 23: Summary of studies excluded from meta-analysis but met criteria for systematic review

Study Author	Stutter group (n)	Control group (n)	Symptom Measure	Stutter group M (SD)	Control group M (SD)	Conclusion
Andrews & Harris (1964)	80	80	General Anxiety Scale for Children (GASC)	Raw scores only*	Raw scores only*	"Anxiety...appears to be evenly distributed between the experimental and the control groups" (pg. 79).
Giorgetti et al. (2015)	32	32	Child Behaviour Checklist (CBCL): <i>anxiety/depression subscale</i>	66.13 (7.25)	56.19 (7.68)	Significant differences on anxiety/depression subscale scores ($p<0.001$)
Tiğrak et al. (2020)	45 [§]	45 [§]	Child Behaviour Checklist (CBCL): <i>anxiety/depression subscale</i>	EC: 5.67 (2.77) MC: 4.53 (3.07) Adol.: 7.8 (4.39)	EC: 0.60 (0.83) MC: 0.67 (0.49) Adol.: 1.6 (2.06)	Significantly higher anxiety/depression subscale scores in the stammering group at across all ages.

Note. *reported raw scores of intergroup comparisons. Outcome of multiple regression analyses reported the correlation between GASC score and stuttering (0.052).

[§] Sample divided into three age groups: early childhood (EC; 0-6 years); middle childhood (MC; 7 – 11 years); adolescence (Adol.; 12 – 18 years). The stuttering and typically developing group were compared on the CBCL in each age group.

Table 24 summarises the three studies excluded in response to recommendation by peer reviewers. The following three studies did not contribute to the summary effect size for the anxiety meta-analysis nor were they included in the qualitative synthesis of depression studies. However, it was possible to calculate the effect sizes for these three studies, which are reported here in the interests of completeness.

Table 24: Summary of studies comprising participants aged between 18 and 25 years, and thereby excluded from the systematic review and meta-analysis

Author	Stutter group	Non-stutter group	Measure	Stutter group mean (SD)	Non-stutter group mean (SD)	Effect size (g) 95% CI
Bray et al. (2003)	21 32 male, 10 female 13 – 19 years	21 Matched for sex 13 – 19 years	Reynolds Adolescent Depression Scale	2.19 (1.12)	1.57 (0.98)	0.58 (-0.04 – 1.20)
Doruk et al. (2008)	26 Male 19 – 23 years	17 Male 19 – 25 years	Beck Anxiety Inventory	19 (15.5)	5.5 (2.1)	1.09 (0.44 – 1.74)
			Beck Depression Inventory	18.2 (11.2)	4.1 (1.9)	1.57 (0.87 – 2.26)

Appendix E: Score distributions by group

Violin plots showing distribution of scores for child- and parent-reported subscale scores by group. The width of the plot indicates how frequently a value occurs in a dataset (wider = values occur more frequently), i.e. density. The 'tails' of the plot represent the density of more extreme values in the distribution. When the 'tail' of the violin plot extends beyond the whiskers of the boxplot, this may indicate outlier scores.

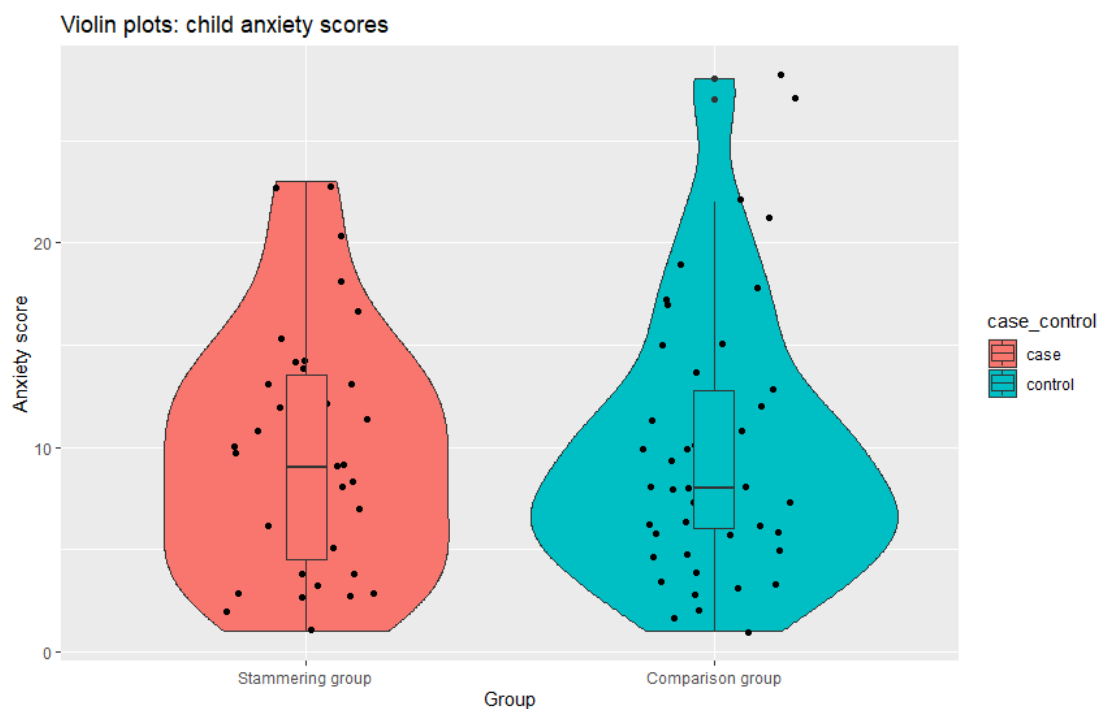


Figure 13: Distribution of child-reported anxiety scores by group

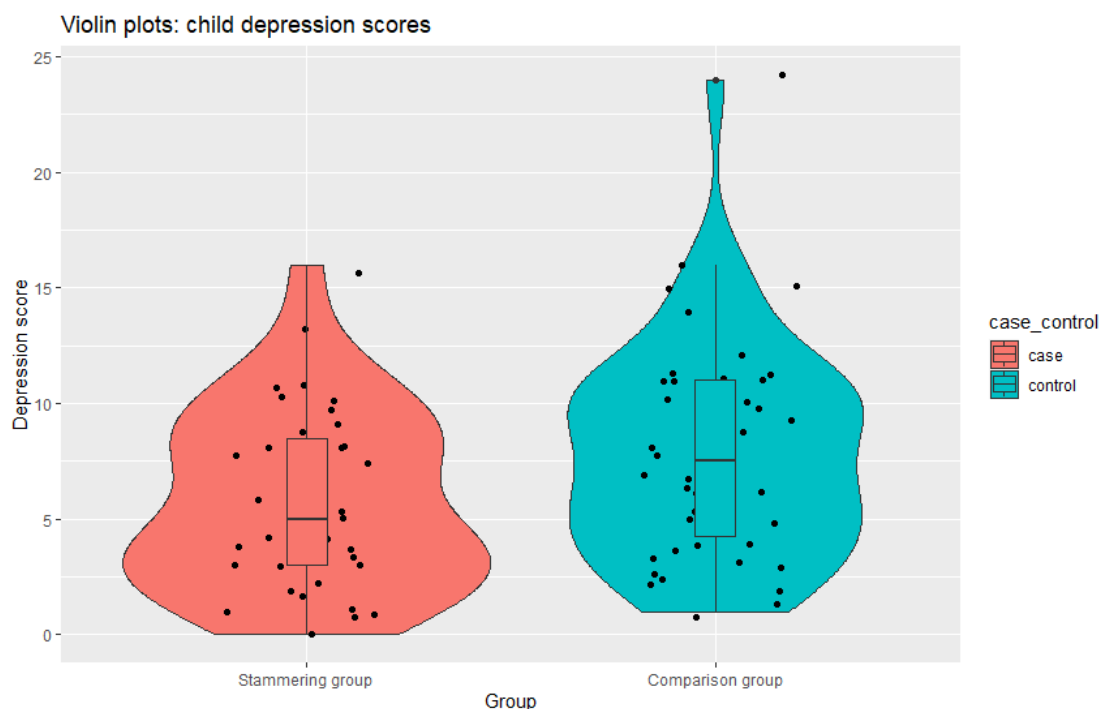


Figure 14: Distribution of child-reported depression scores by group

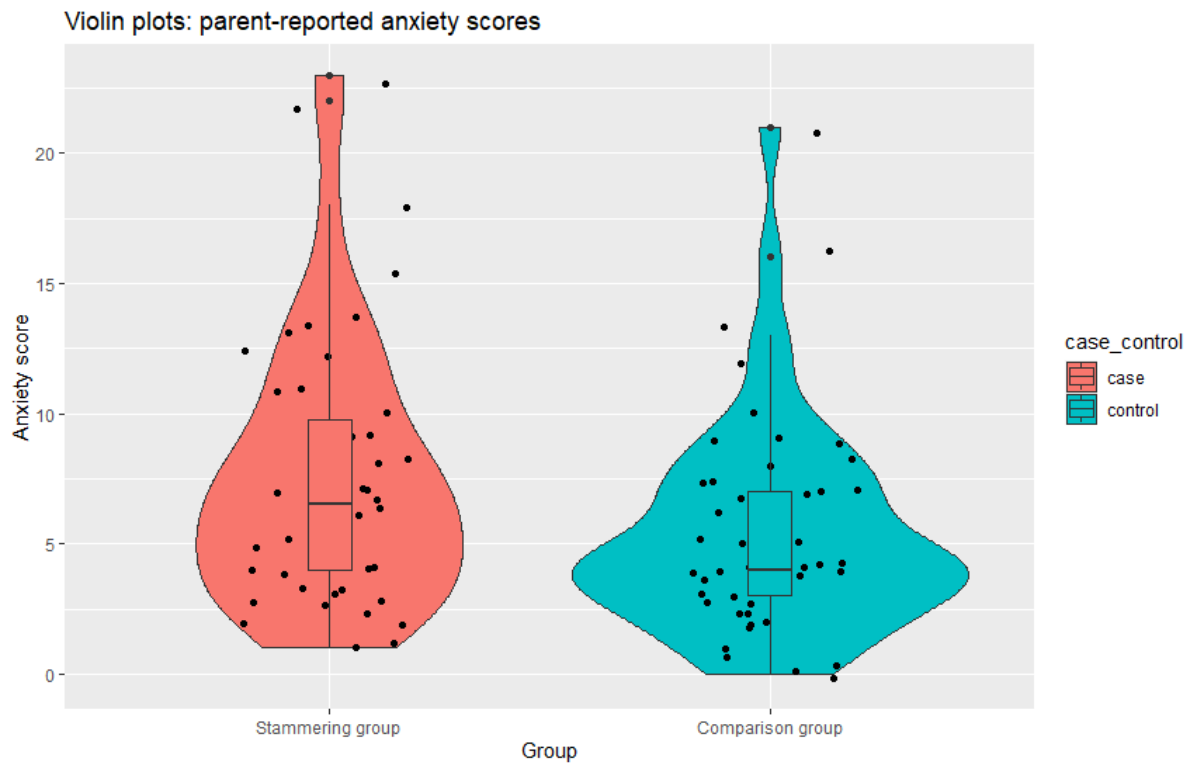


Figure 15: Distribution of parent-reported anxiety scores by group

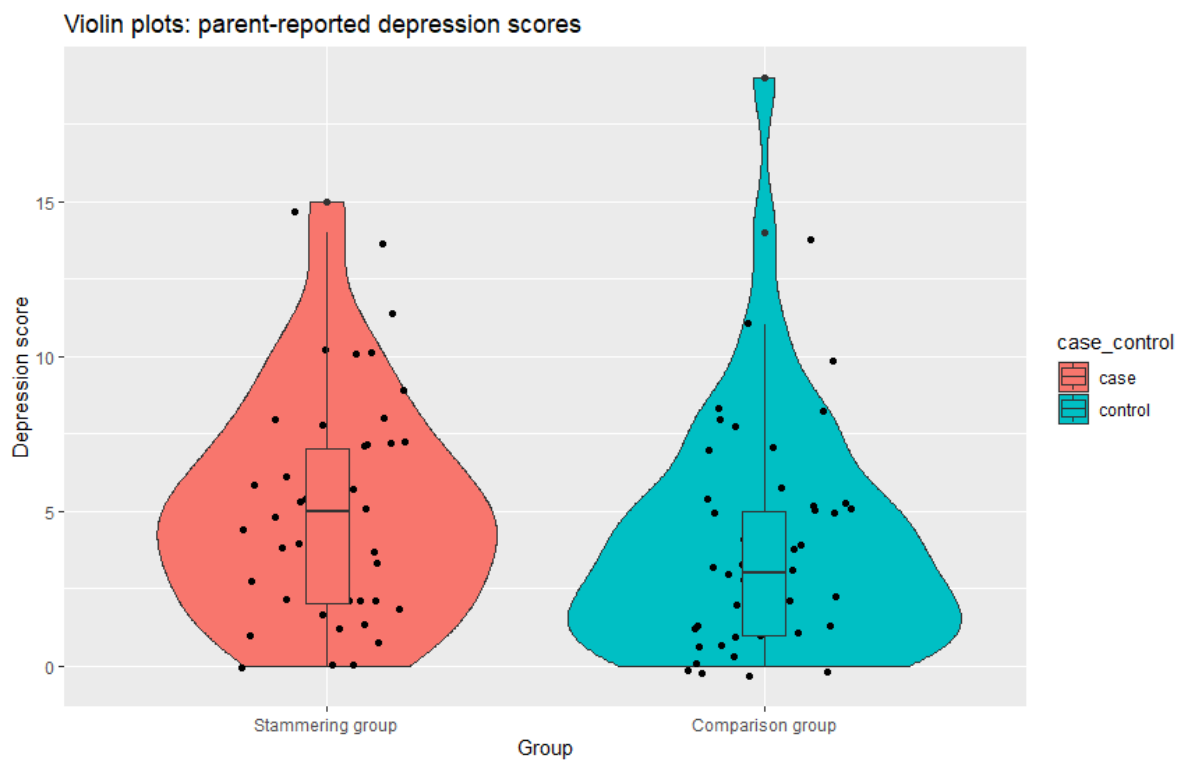


Figure 16: Distribution of parent-reported depression scores by group

Appendix F: MCS Variables

The wording of the speech, language and communication question that formed the basis of the two binary grouping variables (“stuttering” and “speech & language problems”). This information is readily available in the MCS documentation, which is accessible via the UK Data Service. Readers are also directed to the author’s pre-registration: <https://osf.io/wfk5h>

MCS2 (age 3)

University of London, Institute of Education, Centre for Longitudinal Studies.

(2023). *Millennium Cohort Study: Age 3, Sweep 2, 2004*. [data collection]. 11th Edition. UK Data Service. SN: 5350, DOI: <http://doi.org/10.5255/UKDA-SN-5350-6>

Do you have any concerns about [child’s] speech and language? IF YES:
What are your concerns?

- 1 No concerns
- 2 His/her language is developing slowly
- 3 S/he doesn't seem to understand other people
- 4 S/he pronounces words poorly
- 5 S/he doesn't hear well
- 6 S/he stutters
- 7 Other

MCS3 (age 5)

University of London, Institute of Education, Centre for Longitudinal Studies.

(2022). *Millennium Cohort Study: Age 5, Sweep 3, 2006*. [data collection]. 9th Edition. UK Data Service. SN: 5795, DOI: <http://doi.org/10.5255/UKDA-SN-5795-6>

Do you have any concerns about [^Cohort child's name]'s speech and language?

- 1 No concerns
- 2 His/her language is developing slowly
- 3 S/he doesn't seem to understand other people
- 4 S/he pronounces words poorly
- 5 S/he doesn't hear well

6 S/he stutters
95 Other (specify)

MCS5 (age 11)

University of London, Institute of Education, Centre for Longitudinal Studies.

(2022). *Millennium Cohort Study: Age 11, Sweep 5, 2012*. [data collection]. 6th Edition. UK Data Service. SN: 7464, DOI: <http://doi.org/10.5255/UKDA-SN-7464-6>

Does [^Cohort child's name] have any problems with any of the following...?

- 1 ...a stammer or stutter?
- 2 ...another problem with talking?
- 3 ...a problem with understanding what people say? None of these

MCS6 (age 14)

University of London, Institute of Education, Centre for Longitudinal Studies.

(2023). *Millennium Cohort Study: Age 14, Sweep 6, 2015*. [data collection]. 7th Edition. UK Data Service. SN: 8156, DOI: <http://doi.org/10.5255/UKDA-SN-8156-7>

Has a doctor or other health professional EVER told you that [^Cohort member's name] has any problems with any of the following?

- 1a stammer or stutter?
- 2another problem with talking?
- 3a problem with understanding what people say?
- 4 None of these

If yes to any of the above, ask the following question for each problem reported:

- b) Does cohort member have [problem]...
- ...now?
 - ...not now, but had it in the past?

Table 25: Proportion of missing data for each variable

Variable	Missing data (n)	Missing data (%)
Covariates		
Child sex	2	0.01%
Child ethnicity	100	0.58%
OECD 60% median poverty	50	0.29%
OECD weighted income quintile	50	0.29%
Mother NVQ equivalent	3272	19%
Anx/dep diagnosis (mother)	32	1.85%
Anx/dep diagnosis (father)	2695	15.62%
Malaise Inventory MCS1 (mother) ¹¹	1232	7.14%
Kessler MCS2 (mother)	3971	23%
Kessler MCS3 (mother)	3229	18.71%
Kessler MCS4 (mother)	4538	26.3%
Kessler MCS5 (mother)	5291	30.66%
Kessler MCS6 (mother)	6851	39.70%
Kessler MCS7 (mother)	10,063	58.32%
Malaise Inventory MCS1 (father)	5560	32.22%
Kessler MCS2 (father)	7511	43.53%
Kessler MCS3 (father)	7486	43.38%
Kessler MCS4 (father)	8775	50.85%
Kessler MCS5 (father)	9448	54.75%
Kessler MCS6 (father)	10,817	62.69%
Kessler MCS7 (father)	12,689	73.53%
ASD	992	5.75%
ADHD	992	5.75%
Ever bullied	2057	11.92%
BAS t-scores	2715	15.73%
Bracken school readiness std score	3435	19.91%
Outcome variables		
Emotion subscale (mcs2)	2723	15.78%
Emotion subscale (mcs3)	2770	16.05%
Emotion subscale (mcs4)	4088	23.69%
Emotion subscale (mcs5)	4605	26.69%
Emotion subscale (mcs6)	6585	38.16%
Emotion subscale (mcs7)	9138	52.96%
Emotion MCS7 (self-report)	7528	43.63%
Peer Problem MCS7 (self-report)	7529	43.63%
SMFQ	6119	35.46%
Kessler-6	7300	42.30%
Anxiety/depression diagnosis	7308	42.35%
Self-harm (mcs6)	6114	35.43%
Self-harm (mcs7)	7592	44%
Suicide attempt (mcs7)	7547	43.74%

¹¹ In MCS1, parents (mother and father) completed the 9-item Rutter Malaise Inventory, which assesses anxiety, depression and somatic symptoms (see Johnson Atkinson & Rosenberg, 2015). Data from this scale were not included in the present analyses but are reported in the appendices for completeness.

Appendix G: Summary Statistics

Table 26: Summary statistics for categorical variables by group

Variable	Stutter only (n=296)	Speech/language problem only (n=2,871)	Stutter & speech/language problems (n=269)	No reported problems (n=13,820)
Child sex				
Male	191 (64.5%)	1,830 (63.7%)	204 (75.8%)	6,597 (47.7%)
Female	105 (35.5%)	1,041 (36.3%)	65 (24.2%)	7,223 (52.3%)
Child ethnic group				
White	249 (84%)	2,404 (83.7%)	226 (84%)	11,321 (81.9%)
Black	10 (3.4%)	97 (3.4%)	4 (1.5%)	529 (3.8%)
Indian	5 (1.7%)	49 (1.7%)	3 (1%)	390 (2.8%)
Mixed	12 (4.1%)	78 (2.7%)	9 (3.3%)	426 (3.1%)
other	4 (1.4%)	48 (1.7%)	3 (1%)	196 (1.4%)
Pakistani & Bangladeshi	16 (5.4%)	195 (6.8%)	24 (8.9%)	958 (6.9%)
Socio-economic status: OECD 60% poverty threshold				
Above	186 (62.8%)	1,680 (58.5%)	145 (53.9%)	9,183 (66.4%)
Below	110 (37.2%)	1,191 (41.5%)	124 (46.1%)	4,637 (33.6%)
Socio-economic status: OECD Income quintile				
Lowest	74 (25%)	833 (29%)	94 (35.7%)	3,052 (22.1%)
Second	67 (22.6%)	676 (23.5%)	61 (22.7%)	3,068 (22.2%)
Third	59 (19.9%)	568 (19.8%)	42 (15.6%)	2,666 (19.3%)
Fourth	46 (15.5%)	450 (15.7%)	40 (14.9%)	2,571 (18.6%)
Highest	50 (16.9%)	344 (12%)	32 (11.9%)	2,463 (17.8%)
Socio-economic status: mother NVQ-equivalent				
Overseas qualification	15 (5.1%)	146 (5.1%)	20 (7.4%)	745 (5.4%)
Level 1	46 (15.5%)	514 (17.9%)	65 (24.2%)	1885 (13.6%)
Level 2	130 (43.9%)	1,336 (46.5%)	125 (46.5%)	5901 (42.7%)

Level 3	23 (7.8%)	237 (8.3%)	21 (7.8%)	1,427 (10.3%)
Level 4	69 (23.3%)	561 (19.5%)	32 (11.9%)	3,340 (24.2%)
Level 5	13 (4.4%)	77 (2.7%)	6 (2.2%)	522 (3.8%)
ASD diagnosis				
No	291 (98.3%)	2,510 (87.4%)	211 (78.4%)	13,623 (98.6%)
Yes	5 (1.7%)	361 (12.6%)	58 (21.6%)	197 (1.4%)
ADHD diagnosis				
No	287 (97%)	2,648 (92.2%)	224 (83.3%)	13,571 (98.2%)
Yes	9 (3%)	223 (7.8%)	45 (16.7%)	249 (1.08%)
Ever bullied (self-report)				
No	55 (18.6%)	659 (23%)	65 (24.2%)	3,145 (22.8%)
Yes	241 (81.4%)	2,212 (77%)	204 (75.8%)	10,675 (77.2%)
Mother ever diagnosed with serious anxiety or depression				
No	147 (49.7%)	1,280 (44.6%)	97 (36.1%)	7,581 (54.9%)
Yes	149 (50.3%)	1,591 (55.4%)	172 (63.9%)	6,239 (45.1%)
Father ever diagnosed with serious anxiety or depression				
No	230 (77.7%)	2,270 (79.1%)	189 (70.3%)	11,317 (81.9%)
Yes	66 (22.3%)	601 (20.9%)	80 (29.7%)	2,503 (18.1%)

Note: Ns unweighted. Estimates based on more frequent category assigned across the five imputed datasets.

Table 27: Weighted descriptive statistics for observed (unimputed) data

	Stuttering Group	Speech & language problems	Stutter and Speech & language problems	No difficulties
	Mean (SE)	Mean (SE)	Mean (SE)	Mean (SE)
SDQ internalising scores				
MCS2	2.84 (0.28)	3.19 (0.10)	3.26 (0.29)	2.36 (0.04)
MCS3	2.27 (0.23)	3.07 (0.11)	3.72 (0.44)	2.01 (0.04)
MCS4	2.50 (0.25)	3.41 (0.12)	3.51 (0.36)	2.18 (0.04)
MCS5	3.51 (0.38)	4.40 (0.15)	4.70 (0.43)	2.60 (0.05)
MCS6	3.68 (0.42)	5.10 (0.16)	5.30 (0.46)	3.06 (0.05)
MCS7	3.52 (0.44)	4.91 (0.16)	4.91 (0.44)	3.28 (0.06)
Self-report measures				
SMFQ (MCS6)	6.06 (0.63)	5.90 (0.18)	5.54 (0.53)	5.58 (0.08)
Kessler-6 (MCS7)	7.08 (0.45)	7.42 (0.15)	7.27 (0.55)	7.25 (0.07)
SDQ internalising (MCS7)	5.83 (0.33)	5.98 (0.11)	5.65 (0.36)	5.55 (0.05)

Appendix H: LGCMs

Table 28: Results of LGCM 1 (baseline model) showing mean intercept and slope for internalising symptoms for the whole sample

Parameter	Estimate	SE	df	p-value
Fixed Effects				
Intercept	2.50	0.02	1375.30	<.001
Linear Slope	0.09	.003	329.72	<.001
Random Effects (variance)				
Intercept	3.55	0.10	593.66	<.001
Linear Slope	0.04	0.001	73.96	<.001

Note – model fit indices CFI = 0.94, TLI = 0.94, RMSEA = 0.066, SRMR = 0.07.

Table 29: Regression statistics for conditional model (LGCM 2) with two time-invariant predictors (grouping variables).

		Unadjusted model				Adjusted model*			
Predictor	Parameter	Estimate	SE	df	p-value	Estimate	SE	df	p-value
Stutter	Intercept	0.45	0.15	439.00	.003	0.263	0.15	306.553	.08
	Linear Slope	0.01	0.02	31.19	.648	0.006	0.02	31.466	.74
Speech & language problems	Intercept	1.05	0.07	3284.43	<.001	0.486	0.069	260.264	<.001
	Linear Slope	0.06	0.01	61.31	<.001	0.025	0.008	73.826	.004

Note – model indices for unadjusted model (CFI = 0.942, TLI = 0.934, RMSEA = 0.052, SRMR = 0.05); and for adjusted model (CFI = 0.963, TLI = 0.953, RMSEA = 0.02, SRMR = 0.03). *Covariates included in the adjusted model comprised time-invariant variables: child sex, child ethnicity, SES (3 measures), cognitive ability (verbal and non-verbal ability); and time-varying variables: mother and father Kessler-6 scores at MCS2 through to MCS7.

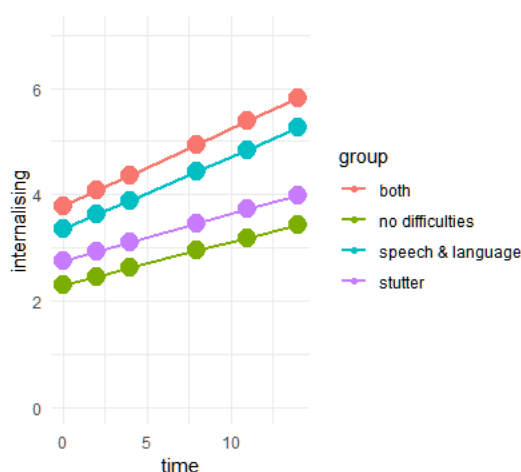


Figure 17: Linear internalising symptom trajectories for all four groups (unadjusted model)

Appendix I: univariate models

Table 30: Results of univariate logistic regression models examining effect of individual predictors on likelihood of scoring above threshold on the SDQ Emotion Subscale (parent-report)

Predictor	OR	df	p-value	Lower CI	Upper CI
(Intercept)	0.19	111.58	<.001	0.13	0.26
Child sex (female)	1.82	466.29	.02	1.09	3.04
(Intercept)	0.16	31.61	<.001	0.10	0.25
OECD poverty (below)	2.36	70.63	.003	1.34	4.13
(Intercept)	0.14	43.48	<.001	0.08	0.22
Mother anxiety or depression	2.35	102.55	.003	1.34	4.13
(Intercept)	0.21	65.02	<.001	0.15	0.29
Father anxiety or depression	1.45	35.94	.25	0.76	2.75
(Intercept)	0.18	37.24	<.001	0.12	0.25
ASD diagnosis	5.62	106.64	<.001	2.79	11.33
(Intercept)	0.20	25.98	<.001	0.14	0.28
ADHD diagnosis	3.30	31.15	.01	1.37	7.97
(Intercept)	0.16	48.13	<.001	0.10	0.25
Co-occurring SLCN	2.09	25.36	.03	1.07	4.06
(Intercept)	0.22	10.03	.005	0.08	0.55
Ever bullied	1.07	13.22	.87	0.42	2.75

Table 31: Results of univariate logistic regression models examining the effect of individual predictors on scoring above threshold on the SMFQ

Predictor	OR	df	p-value	Lower CI	Upper CI
(Intercept)	0.11	41.36	<.001	0.07	0.18
Child sex (female)	4.20	68.64	<.001	2.23	7.88
(Intercept)	0.16	91.95	<.001	0.10	0.23
OECD poverty (below)	1.87	70.60	.04	1.02	3.41
(Intercept)	0.15	74.05	<.001	0.09	0.25
Mother anxiety/ depression	1.61	73.59	.14	0.86	3.01
(Intercept)	0.18	26.45	<.001	0.12	0.27
Father anxiety/ depression	1.45	17.13	.36	0.63	3.31
(Intercept)	0.20	204.12	<.001	0.15	0.26
ASD diagnosis	1.04	35.61	.93	0.36	3.03
(Intercept)	0.20	117.96	<.001	0.15	0.27
ADHD diagnosis	0.97	54.68	.96	0.32	2.91
(Intercept)	0.22	452.02	<.001	0.15	0.31
Co-occurring SLCN	0.81	297.36	.44	0.46	1.40
(Intercept)	0.06	34.93	<.001	0.02	0.18
Ever bullied (yes)	3.36	28.65	.05	0.99	11.46

Table 32: Results of univariate logistic regression models examining the effect of individual predictors on scoring above threshold on SDQ emotion subscale (self-report)

Predictor	OR	df	p-value	Lower CI	Upper CI
(Intercept)	0.06	196.69	<.001	0.04	0.11
Child sex (female)	5.80	196.18	.001	2.98	11.30
(Intercept)	0.12	131.88	<.001	0.08	0.19
OECD poverty (below)	1.52	44.07	.24	0.75	3.08
(Intercept)	0.07	210.06	<.001	0.04	0.13
Mother anxiety or depression	2.75	170.06	.005	1.37	5.50
(Intercept)	0.14	32.12	<.001	0.09	0.22
Father anxiety or depression	1.02	18.89	.97	0.40	2.57
(Intercept)	0.13	29.56	<.001	0.09	0.20
ASD diagnosis	1.32	20.44	.64	0.40	4.40
(Intercept)	0.14	38.31	<.001	0.10	0.21
ADHD diagnosis	0.76	14.32	.75	0.12	4.69
(Intercept)	0.17	26.42	<.001	0.10	0.28
Co-occurring SLCN	0.61	36.16	.19	0.29	1.30
(Intercept)	0.07	24.08	<.001	0.02	0.20
Ever bullied (yes)	2.35	17.78	.18	0.66	8.40

Table 33: Results of univariate logistic regression models estimating size of the effect of individual predictors on scoring above threshold on the Kessler-6

Predictor	OR	df	p-value	Lower CI	Upper CI
(Intercept)	0.10	25.86	<.001	0.06	0.17
Child Sex (female)	3.41	45.37	0.001	1.71	6.81
(Intercept)	0.13	61.46	<.001	0.08	0.20
OECD poverty (below)	2.00	165.89	0.02	1.10	3.67
(Intercept)	0.08	75.06	<.001	0.04	0.15
Mother anxiety or Depression	3.08	273.66	0.001	1.58	6.01
(Intercept)	0.13	30.97	<.001	0.08	0.21
Father anxiety or depression	1.96	106.63	0.04	1.03	3.75
(Intercept)	0.15	43.34	<.001	0.11	0.22
ASD diagnosis	1.82	16.22	0.31	0.55	6.01
(Intercept)	0.16	41.17	<.001	0.11	0.23
ADHD diagnosis	1.47	15.94	0.55	0.38	5.66
(Intercept)	0.16	48.90	<.001	0.10	0.25
Co-occurring SLCN	1.11	353.85	0.73	0.62	1.97
(Intercept)	0.06	34.93	<.001	0.02	0.18
Ever bullied (yes)	3.36	28.65	0.05	0.99	11.46

Table 34: Results of univariate logistic regression analyses examining the effect of individual predictors on being diagnosed with anxiety or depression aged 17

Predictors	OR	df	p-value	Lower CI	Upper CI
(Intercept)	0.09	17.70	<.001	0.05	0.16
Child sex (female)	2.78	33.34	.01	1.26	6.12
(Intercept)	0.10	229.71	<.001	0.07	0.16
OECD poverty (below)	1.73	124.79	.11	0.88	3.40
(Intercept)	0.07	346.46	<.001	0.04	0.13
Mother anxiety or depression	2.50	100.77	.02	1.17	5.31
(Intercept)	0.10	21.64	<.001	0.06	0.18
Father anxiety or depression	1.93	63.65	.08	0.92	4.03
(Intercept)	0.12	164.61	<.001	0.09	0.18
ASD diagnosis	1.50	21.86	.50	0.45	5.03
(Intercept)	0.13	77.67	<.001	0.09	0.18
ADHD diagnosis	1.33	24.57	.67	0.34	5.20
(Intercept)	0.13	141.61	<.001	0.08	0.20
Co-occurring SLCN	1.03	28.43	.94	0.46	2.30
(Intercept)	0.03	9.86	.003	0.005	0.24
Ever bullied	4.60	10.35	.12	0.63	33.57