A mediation model of social anxiety development during early childhood stuttering

Mark Onslow[a], Brett Dyer[b], Mark Jones[c], Robyn Lowe[a], Sue O'Brian[a], and Ross Menzies[a]

[a] University of Technology Sydney, Australian Stuttering Research Centre, NSW, Australia

[b] Griffith University, Griffith Biostatistics Unit, Griffith Health, QLD, Australia

[c] Bond University, Faculty of Health Sciences & Medicine, QLD, Australia

[*] Corresponding Author: Mark Onslow, Australian Stuttering Research Centre, University of Technology Sydney, Building 1, 15 Broadway, Ultimo, NSW, 2007, AUSTRALIA

e-mail: Mark.Onslow@uts.edu.au

Keywords: Stuttering, childhood, anxiety

Conflict of Interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing.

Abstract

Purpose: Stuttering is associated with clinically significant social anxiety, which emerges during early childhood for some, but not all, children who begin to stutter. The purpose of this paper is to develop a model of social anxiety development during early childhood stuttering and to present an empirical method by which it can be tested.

Method: We propose a mediation model of how the exposure variable of stuttering may lead to an outcome of social anxiety. Our model includes confounder and mediator variables. We explain the concepts and procedures of mediation analysis and present a method to test our model.

Results: We present the idea that negative peer responses to stuttering and negative self-perception of children are mediators of social anxiety development. We propose several confounder variables that involve children, their parents, and the home environment. We depict our model with a Directed Acyclic Graph, and we present details of how it can be tested with a longitudinal research design.

Discussion: This is the first attempt to model the development of social anxiety shortly after stuttering onset with an empirically testable method. The intended benefit of this innovation is to direct future clinical directions for the clinical management of stuttering arising shortly after childhood onset.

Stuttering and social anxiety

During the past decades, it has become apparent from a large body of independent reports that stuttering is associated with social anxiety (Chu et al., 2020; Craig & Tran; 2006; Gabel et al., 2002; Kraaimaat et al 2002; Tomisato et al., 2022; Tran et al., 2011). A meta-analysis showed this to be the case for state and trait anxiety (Craig & Tran, 2014). Those who stutter are particularly at risk for social anxiety disorder (Blumgart et al., 2010; Iverach et al., 2009). The population prevalence for such a diagnosis is 8–13% (Kessler et al. 2005, Kikuchi et al., 2023), yet, based on cases presenting to clinics, the prevalence is much higher for stuttering (Blumgart et al., 2010; Stein et al., 1996), with one report showing 34-fold increased odds for stuttering (Iverach et al. 2009). The quality of life and burden of illness impairment associated with stuttering is equivalent to that of chronic health conditions such as diabetes, cardiovascular disease, and cancer (Norman et al., 2023), and social anxiety is likely to contribute to those impacts on life. During adulthood, around half of adults who seek clinical services for stuttering wish to deal with their social anxiety (Vanryckeghem & Van Eerdenbrugh, 2024; Yaruss et al. 2002).

Not surprisingly, these issues develop early in life. For social anxiety in general, a systematic review and meta-analysis concluded that children and adolescents who stutter present with increased anxiety symptoms in comparison to their peers who do not stutter (Bernard et al., 2022). One report indicated that 24% of children 7–12 years old who stutter are diagnosed with social anxiety disorder (Iverach et al., 2016). There are some reports of anxiety in pre-school children. A community cohort study included 173 three-year-olds who stuttered, and concluded “that early social, emotional and behavioral difficulties may be apparent in children who stutter as young as 3 years old” (McAllister, 2016, p. 30). Another cohort study (Briley et al. 2019) found significant anxiety differences between 4-year-olds whose parents reported stuttering and those whose parents did not. A small study of stuttering

children reported differences from control children at 3–6 years (Tığrak et al., 2020).

Establishing a model of social anxiety development during early childhood

It is clear that anxiety as a clinically important feature of the disorder emerges during the early years of life for many—but not all—children who stutter. Why this occurs is of clinical and theoretical interest. Theoretical modelling of the process by which this early anxiety development occurs, and its subsequent empirical verification, could generate benefits by directing future clinical research, as described later in this paper. There are numerous models of social anxiety maintenance for the general population (e.g., Clark & Wells, 1995; Hartman, 1983; Hofman, 2007; Moscovitch, 2009; Rapee & Heimberg, 1997; Schlenker & Leary, 1982; Spence & Rapee, 2016) and one has been devised specifically for stuttering (Iverach et al., 2017).

In general, these models of anxiety maintenance propose that high expectations of performance in social situations and perceived incapacity to meet those expectations leads to fear of negative evaluations from others. Within feared situations, the person becomes hypervigilant to negative threat cues, including physiological arousal associated with anxiety, negative cognitions, and negative self-evaluations. This self-focused attention affects the ability to observe and process disconfirming social cues. To avoid negative self-evaluations from others and reduce anxiety, safety behaviors are used. However, self-focused attention and safety behaviors can exacerbate anxiety and lead to the feared outcome of poor performance, thereby, confirming fears. Further, these negative responses to social situations can reduce attentional resources being available to attend to the situation, increasing the risk that the feared outcome of negative evaluations from others will in fact occur. These processes form a feedback cycle that maintains, or even exacerbates, anxiety.

There is evidence that the models of social anxiety maintenance apply to chronic stuttering in adults (Lowe, Menzies, et al., 2021). One model of anxiety maintenance is specific to stuttering; Iverach et al. (2017), drawing on the Clark and Wells (1995) and Rapee & Heimberg (1997) models, incorporated interrelated factors responsible for anxiety maintenance with adults who

stutter. This model included both self-focused attention and externally focused attention to negative social threat cues. Iverach et al. suggested that anxiety maintenance with stuttering is unique and complex due to having a real condition from which fears arise. For example, negative self-perceptions about speaking in social settings can be justified if negative experiences have occurred. It is the case that many people who stutter experience negative reactions from others, and these negative experiences can occur early in life, as noted above.

Overall, these models primarily explain anxiety maintenance during adulthood and do not focus on the origins of that anxiety. As such, they do not inform the development of a model to explain why some children who stutter develop anxiety and some do not. The Spence and Rapee (2016) model does present such a comprehensive explanation, implicating an interplay between moderating and mediating variables in early and adult life: gender, age, genetics, temperament, culture, parent and peer influence, and aversive social outcomes. Hence, the present model builds on that material to develop a stuttering-specific model of anxiety development during childhood.

The concept of mediation

The proposed mechanism of stuttering causing social anxiety can be thought of with a *mediation model*. The value of mediation arises when there is interest in explaining the mechanism of how an exposure *E* causes an outcome *O*. An exposure could be any variable, such as a behavior, health condition, or environmental feature. If there is reason to believe that part of the reason an exposure *E* causes outcome *O* is because of an intermediate variable *M*, that variable *M* is called a mediator.

A Directed Acyclic Graph (DAG) (Greenland & Brumback, 2002; Greenland et al., 1999) can depict such a relationship, using arrows to reflect one variable potentially causing another. A DAG presents variables starting with those that occurred earliest in a temporal sequence and ending with those that occurred last. The characteristics of a DAG are a flow towards the outcome *O* in one direction only, without any cycles (“loops”) within it. Figure 1 demonstrates a simple mediation model of an exposure *E* causing an outcome *O*, and a

mediator *M*. The pathway $E \rightarrow M \rightarrow O$ is called the indirect effect, which includes some portion of the effect of exposure E on outcome *O* being due to the mediator *M*. The other pathway $E \rightarrow O$ is the direct effect which is not related to the mediator *M*.

INSERT FIGURE 1 AROUND HERE

The process of splitting the effect of exposure *E* and outcome *O* into a direct pathway and an indirect pathway is called *effect decomposition* (Rijnhart et al., 2021) and can explain the mechanism by which the outcome develops. In healthcare, effect decomposition can identify potential interventions to prevent the onset of the outcome in question. For example, if it is found that a mediator *M* substantially explains the causal effect of exposure *E* on outcome *O*, an intervention for mediator *M* could be targeted to reduce the risk of developing outcome *O*. This is of particular interest when the mediator *M* is clinically modifiable.

Causal mediation analysis

Traditional approaches to mediation analysis such as the difference method (MacKinnon et al., 2012; VanderWeele, 2016) and the product method (Alwin & Hauser, 1975; Baron & Kenny, 1986; MacKinnon et al., 2012; VanderWeele, 2016) are simplistic and often will not isolate the desired causal effects. Instead, a *causal mediation analysis* approach can be utilized to estimate unbiased causal effects (VanderWeele, 2016). The key factor that separates the causal mediation analysis approach from traditional approaches is the assumptions about confounding variables that underpin the analysis (VanderWeele, 2016). When considering whether an exposure *E* causes an outcome *O*, a confounder *C* is a variable that causes *E* and also causes *O*. This is shown in Figure 2.

INSERT FIGURE 2 AROUND HERE

A simple, commonly used, hypothetical example of a confounding variable would be in an empirical study of the relationship between ice-cream sales and shark attacks. Ice-cream sales and shark attacks are correlated, but to determine any causal association, it is necessary to account for the weather as a confounder. If it is a hot day, then more people will be at the beach, so ice cream sales will be high. Also, if it is a hot day, then more people will be swimming, and the probability of a shark attack will be higher. If the confounder of the weather is adjusted for in the analysis, then ice-cream sales will no longer be associated with shark attacks, and the conclusion is that ice-cream sales do not cause shark attacks.

Assumptions for causal mediation analysis

In a mediation analysis, the mediating variable *M* (see Figure 1) is incorporated into the analysis. Mediation analysis requires the estimation of the effect of (a) the exposure on the outcome, (b) the mediator on the outcome, and (c) the exposure on the mediator. Because of this, analysis requires four assumptions about confounding, so that each of the three effects are unbiased. Any empirical verification of a causal mediation model is underpinned by the following four assumptions.

Assumption 1: No unmeasured exposure-outcome confounder

Adjustment should be made for all confounders C of the exposure and the outcome.

Assumption 2: No unmeasured mediator-outcome confounder

Estimation of the indirect effect $E \rightarrow M \rightarrow O$ requires the estimation of the mediator on the outcome. For this effect to be unbiased it is necessary to adjust for confounders C of the mediator *M* and outcome *O*, as shown by *C* in Figure 3. Hence, it is assumed that all such relevant confounders C have been measured.

INSERT FIGURE 3 AROUND HERE

147

148 **Assumption 3: No unmeasured exposure-mediator confounder**

149 With the same logic as Assumption 2, an unbiased estimation of the indirect effect $E \rightarrow$
150 $M \rightarrow O$ requires adjustment for confounders C of the mediator M and exposure E, as shown
151 by C in Figure 4. Hence, it is assumed that all such relevant confounders C have been
152 measured.

153

154 INSERT FIGURE 4 AROUND HERE

155

156 **Assumption 4: No mediator-outcome confounder is caused by the exposure**

157 Figure 5 shows two ways that a mediator-outcome confounder C can itself be
158 influenced by the exposure E. This can occur as part of the direct effect $E \rightarrow C \rightarrow O$ or as part
159 of the indirect effect $E \rightarrow C \rightarrow M \rightarrow O$. It is necessary to adjust for confounder C in the
160 analysis to satisfy Assumption 2; however, doing so blocks the two causal pathways, $E \rightarrow C$
161 $\rightarrow O$ and $E \rightarrow C \rightarrow M \rightarrow O$, consequently introducing bias. Hence, a required assumption is
162 that no mediator-outcome confounder is caused by the exposure.

163

164 INSERT FIGURE 5 AROUND HERE

165

166 **A DAG for the development of social anxiety with early stuttering**

167 DAGs are helpful tools for communicating assumptions or hypotheses about causal
168 processes. They can also help to identify confounders so that researchers can attempt to avoid
169 violations of assumptions 1–4. Figure 6 contains the DAG summarising our knowledge and
170 hypotheses about the process of how stuttering may lead to social anxiety early in life through
171 negative peer reactions and negative self-perception. This DAG in Figure 6 was constructed

according to the Tennant et al. (2021) guidelines.

INSERT FIGURE 6 AROUND HERE

Arranging variables in order of occurrence

Firstly, variables were arranged in time order, starting with those that were set earliest and ending with those that happened last. It is well documented that around two-thirds of those who stutter have the variable *family history of stuttering* (Bloodstein et al., 2021), and this history exists prior to the child's birth. This variable was included in the DAG because of the association between stuttering and anxiety, as described above, raising a possible connection to childhood anxiety. *Parent history of anxiety* is present early in a child's life. It is capable of influencing child anxiety (Song et al., 2022), and it also exists before the child's birth. *Age* and *sex* of the child are set at birth and are both potentially involved in development of childhood anxiety: the chance of anxiety developing will logically increase with age, and the sex of the child contributes to anxiety development, affecting females more often than males (McLean et al, 2011). After birth of the child, the *emotional stability of the home* may be affected. This might occur, for example, with postpartum depression, which may then affect the child's anxiety level (Slomian et al., 2019). There is evidence that there is no difference in anxiety-prone temperament between children who stutter and their peers who do not stutter (Kefalianos et al., 2014). However, a child who has an anxiety-prone temperament is more likely to develop or experience anxiety after commencing stuttering than a child without such a temperament. This is a logical prospect that applies to any child, regardless of stuttering status.

The exposure of *stuttering* then occurs. Then the mediators of *negative peer reactions* and *negative self-perception*, and the outcome of *social anxiety* are ordered as we hypothesize

in the mediation model. There is evidence implicating negative peer reactions in the development of social anxiety for children who stutter. Peers are aware of stuttering; for example, Ambrose & Yairi (1994) used a research paradigm, which showed that, over a period of 2 years, pre-school children were able to distinguish between stuttering and non-stuttering puppets. Weidner et al. (2015) developed the POSHA-S/Child tool to reveal that 3–7-year-olds had a negative attitude to stuttering. There is direct observational evidence of negative peer reactions to stuttering in pre-school settings; Langevin et al. (2009) video recorded children who stutter in the playground, and three of the four children were shown to experience negative peer reactions to the stuttering: walking away from the child, being ignored or interrupted, and mocking. There is also evidence to implicate negative self-perception in anxiety development with early stuttering. There are recurring parent reports that two-thirds of pre-school children show negative awareness of their early stuttering, with some figures as high as 90% (Langevin et al., 2010). Common parent reports focus on "frustration associated with their stuttering, withdrawal, reduced or changed verbal output, making comments about their inability to talk" (Langevin et al., 2010; p. 407). Negative self-perception has been measured in pre-school children who stutter with the KiddyCAT test of communication attitude for children who stutter (Clark et al., 2012; Węsierska & Vanryckeghem, 2015).

Plausibility of causal effects

The next step of drawing a DAG is to decide which arrows should be included, that is, deciding which variables may cause other variables. It is recommended to only exclude arrows from a DAG where there is strong *a priori* evidence or logic to suggest the effect is non-existent (Tennant et al., 2021). This recommendation arises from the statistical implications of missing confounders. Generally, it is worse to miss a confounder than to adjust for a variable that actually doesn't cause the exposure or the outcome as suspected.

Thus, we started with a saturated DAG, that is, a DAG including all arrows from variables occurring earlier in time leading to variables occurring later in time. We then determined whether there was strong evidence or logic to suggest that an arrow could be excluded. The only arrows that we excluded were (a) from *parent history of anxiety* to *stuttering*, (b) from both *parent history of anxiety* and *family history of stuttering* to *age* and *sex*. Exclusion (a) was based on empirical data showing no evidence of parent anxiety in a clinical cohort of pre-schoolers who stutter (Park et al., 2021). Exclusions (b) were based on logical grounds. To avoid overcrowding the DAG with arrows, we have included "thick arrows" from a variable to indicate arrows to all variables below that occur later in life.

The DAG in Figure 6 implies that *parent history of anxiety* is a mediator-outcome confounder and that *family history of stuttering*, *age*, *sex*, *emotional stability of the home*, and *child temperament* are exposure-mediator, mediator-outcome, and exposure-outcome confounders. Additionally, no mediator-outcome confounder is caused by the exposure. Thus, assuming data are available for the identified confounders, then it is possible to conduct a causal mediation analysis to test this proposed mediation model. That analysis can estimate the indirect effect of stuttering on social anxiety through negative peer reactions and negative self-perception, and the remaining direct effect.

Testing our mediation model of social anxiety development during early childhood

The value of any theoretical position about stuttering is limited by its testability (Packman, 2012). Testing a model establishes its veracity and can lead to its refinement or, ultimately, its abandonment in the face of repeated disconfirmation. To our knowledge, this is the first model about stuttering that is presented along with methodological details about how it might be tested.

Study design

A longitudinal cohort study of stuttering children and control children is the best way to

test the mediation model proposed in this paper. Such a study would recruit children who have recently started stuttering and age- and gender-matched controls who do not stutter, and prospectively follow them. A prospective design allows for the temporal ordering of events to be recorded, as this is an important factor in estimating causal relationships. Stuttering and matched control children could be studied with 4-monthly observations for 3 years after stuttering onset.

A simple recruitment strategy is to source clinics to locate children who have been brought to speech-language pathologists shortly after onset. With this method, when a stuttering child has been recruited from a clinic, a matched nonclinical control can be established. A more methodologically rigorous—and resource-intensive—method is to recruit stuttering and control children from a community sample. Cases of stuttering onset could be identified in the community using previously established methods (Reilly et al., 2013). Those methods included community-distributed fridge magnets describing stuttering, along with a request to contact the researchers if a child starts to stutter.

Measurement of variables.

Parent history of anxiety and *family history of stuttering* could be assessed with parent interview at recruitment. *Child sex* and *child age* would be established with demographic information. The *emotional stability of the home* could be measured at recruitment with the Emotional Stability Scale (Chaturvedi & Chander, 2010) given to all adults living in the child's home. *Child temperament* could be assessed by parents at recruitment with the Children's Behavior Questionnaire (Rothbart et al., 2001).

The exposure variable of *stuttering* could be determined with standard speech-language pathology procedures of parent interview telehealth, supplemented by parents uploading digital video recordings of the child talking during everyday situations. *Negative peer reactions* to stuttering could be measured with a simple method of parent report about

whether they observed that their child had received positive, negative, or neutral responses from peers when they stuttered. A more rigorous method would be to train parents to make observations of the child during everyday situations with peers, siblings, and relatives. With even more methodological rigour, research personnel could make visits to selected everyday child environments and make such observations independently. *Negative self-perception* could be measured at recruitment with the KiddyCat (Vanryckeghem & Brutten, 2007). The outcome variable of *social anxiety* could be measured with the social anxiety subscale of the Spence Children's Anxiety Scale (Spence, 1998).

Statistical analysis

An inverse probability weighting approach coupled with Cox proportional-hazards regression (a survival analysis method) can be used to estimate the direct effect and indirect effect of stuttering (Lange et al., 2012). Inverse probability weighting is a procedure that involves assigning weights to each participant based on the probability of being exposed or unexposed to create a pseudo-population in which the probability of being exposed or unexposed is independent of confounding variables, as is the case in a randomized trial. The inverse probability weighting technique used in mediation analysis extends this idea by using weighting to mimic a sequentially randomized trial in which participants are randomly allocated to either have the exposure or not have the exposure, and then randomly allocated to either have the mediator or not have the mediator, while also taking into account the effect of the exposure on the mediator. This more advanced type of inverse probability weighting accounts for confounding assumptions1–4, assuming the correct specification of the model. Following the inverse probability weighting, the Cox proportional-hazards model can produce unbiased estimates of the indirect effect and direct effect of stuttering. The Cox model also accounts for participation dropout.

A sensitivity analysis for unmeasured confounding can be conducted using E-values.

The E-values indicate the impact that potential unknown/unmeasured confounders would have on the effect estimates (VanderWeele et al., 2017). Thus, the sensitivity analysis would indicate how sensitive the results are to missing confounders.

Potential benefits of a mediation model for social anxiety development during early childhood stuttering

Clearly, early stuttering during childhood is associated somehow with development of social anxiety, manifesting as a core clinical issue during persistent stuttering. An empirically verified model of how social anxiety begins early in life, with mediators and confounders established, would have many benefits and challenges, potentially leading to paradigm shifts with early intervention. Some examples of how this might occur are presented below.

There is consensus in the field of early stuttering intervention that treatment should be delayed for some time after onset rather than beginning immediately (Lowe, Jelčić Jakšic, et al., 2021). Such an approach contains a tacit assumption that the debilitating effects of social anxiety that are associated with stuttering are independent of age. Yet, empirical development of our proposed mediation model may show that assumption to be wrong. Cohort studies report the age of stuttering onset to range from 18 months to 68 months (Ambrose et al., 2015; Reilly et al., 2013). This introduces potential quantitative and qualitative age effects on anxiety development. Quantitatively, some children who begin to stutter will do so earlier than other children, and hence, will be exposed to stuttering for longer than other children, often for a number of years. From a qualitative perspective, the differences in cognitive development between 2-year-olds and 5-year-olds who begin to stutter are substantive. Hence, empirical testing of our model could reveal age to be an exposure-mediator confounder (see Figure 4), or a mediator-outcome confounder (see Figure 5). In other words, it could be that the development of social anxiety indeed is mediated by negative peer reactions or negative self-perception—or both—and that age of onset influences that process.

In such cases, it would not be clinically appropriate to apply a general rule to delay early stuttering intervention.

At present, randomized clinical trials in the field have focused almost exclusively on stuttering severity reduction with early stuttering, using primary outcomes of the observable speech behaviors of stuttering (Sjøstrand et al. 2021). Presumably, this also carries the tacit assumption that stuttering reduction is the ideal way to prevent the development of a range of negative outcomes from stuttering, with social anxiety included. Testing of our model might substantiate this assumption, with the pathway from the exposure of stuttering to the outcome of social anxiety being straightforward, unmediated, and unconfounded by any variables. But, on the other hand, testing of our model might reveal the causal pathway to be affected to a clinically significant extent by child psychological issues. For example, negative self-perception might be a prominent mediator of social anxiety development, with child temperament as a prominent confounder of that mediating variable. In such a case, this would signal potential limitations of interventions designed to control stuttering behaviors, and it would signal a need for continued development of interventions to establish positive self-perception and psychological resilience in stuttering pre-schoolers, as seen in Druker et al. (2019).

Another result to emerge from testing of our model might reveal a prominent role of parents in the development of childhood social anxiety. Regardless of other confounding variables on the pathway from the exposure of stuttering to the outcome of social anxiety, parents may have a clinically important influence. For example, parent history of anxiety, or the emotional stability of adults living in the home (see Figure 6), or both, may be confounders. In such cases, speech pathologists who assess and manage pre-schoolers who stutter may be advised to screen the mental health of all adults in a child's life and provide psychological support services to them as needed.

Conclusion

Anxiety is a core clinical concern for those who stutter and for researchers who seek to understand it. For some children who stutter, social anxiety develops and persists into adulthood, contributing to substantial quality of life impairment and burden of illness. This paper presents the first attempt to explain that process, using a mediation model. Our proposed model involves the exposure variable of stuttering during early childhood and the outcome variable of social anxiety, with several confounder and mediator variables. We present a research method that would enable testing and refinement of this model. The intended benefit of this innovation is to direct future clinical directions for clinical management of stuttering shortly after childhood onset.

Acknowledgments

The authors would also like to acknowledge Damien Liu-Brennan for his scientific copyediting contribution to this publication.

Data availability statement

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

References

Alwin, D. F., & Hauser, R. M. (1975). The decomposition of effects in path analysis. *American Sociological Review*, *40*(1), 37–47. https://doi.org/10.2307/2094445

Ambrose, N. G., & Yairi, E. (1994). The development of awareness of stuttering in preschool children. *Journal of Fluency Disorders*, *19*(4), 229–245. https://doi.org/10.1016/0094-730X(94)90002-7

Ambrose, N. G., Yairi, E., Loucks, T. M., Seery, C. H., & Throneburg, R. (2015). Relation of motor, linguistic and temperament factors in epidemiologic subtypes of persistent and recovered stuttering: Initial findings. *Journal of Fluency Disorders, 45,* 12–26. https://doi.org/10.1016/j.jfludis.2015.05.004

Baron, R. M., & Kenny, D. A. (1986). The moderator–mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. *Journal of Personality and Social Psychology*, *51*(6), 1173–1182. https://doi.org/10.1037/0022-3514.51.6.1173

Bernard, R., Hofslundsengen, H., & Frazier Norbury, C. (2022). Anxiety and depression symptoms in children and adolescents who stutter: A Systematic Review and Meta-Analysis. *Journal of Speech, Language, and Hearing Research*, *65*(2), 624–644. https://doi.org/10.1044/2021_JSLHR-21-00236

Bloodstein, O, Bernstein Ratner, N., & Brundage, S. B. (2021). *A handbook on stuttering* (7th ed.). Plural Publishing.

Blumgart, E., Tran, Y., & Craig, A. (2010). Social anxiety disorder in adults who stutter. *Depression and Anxiety*, *27*(7), 687–692. https://doi.org/10.1002/da.20657

Briley, P. M., O'Brien, K., & Ellis, C. (2019). Behavioral, emotional, and social well-being in children who stutter: Evidence from the National Health Interview Survey. *Journal of Developmental and Physical Disabilities*, *31*(1), 39–53. https://doi.org/10.1007/S10882-018-9625-X

Clark, C. E., Conture, E. G., Frankel, C. B., & Walden, T. A. (2012). Communicative and psychological dimensions of the KiddyCAT. *Journal of Communication Disorders*, *45*(3), 223–234. https://doi.org/10.1016/j.jcomdis.2012.01.002

Clark, D. M. & Wells, A. (1995). A cognitive model of social phobia. In R. G. Heimberg, M. R. Liebowitz, D. A. Hope, & F. R. Schneier (Eds.), *Social phobia: Diagnosis, assessment and treatment*. (pp. 69–93). Guilford Press.

Chaturvedi, M., & Chander, R. (2010). Development of emotional stability scale. *Industrial Psychiatry Journal*, *19*(1), 37–40. https://doi.org/10.4103/0972-6748.77634

Chu, S. Y., Sakai, N., Lee, J., Harrison, E., Tang, K. P., & Mori, K. (2020). Comparison of social anxiety between Japanese adults who stutter and non-stuttering controls. *Journal of Fluency Disorders*, *65*, Article 105767. https://doi.org/10.1016/j.jfludis.2020.105767

Craig, A., & Tran, Y. (2006). Fear of speaking: Chronic anxiety and stammering. *Advances in Psychiatric Treatment*, *12*(1), 63–68. https://doi.org/10.1192/apt.12.1.63

Craig, A., & Tran, Y. (2014). Trait and social anxiety in adults with chronic stuttering: Conclusions following meta-analysis. *Journal of Fluency Disorders*, *40*, 35–43. https://doi.org/10.1016/j.jfludis.2014.01.001

Druker, K. C., Mazzucchelli, T. G., & Beilby, J. M. (2019). An evaluation of an integrated fluency and resilience program for early developmental stuttering disorders. *Journal of Communication Disorders, 78,* 69–83. https://doi.org/10.1016/j.jcomdis.2019.02.002

Gabel, R. M., Colcord, R. D., & Petrosino, L. (2002). Self-reported anxiety of adults who do and do not stutter. *Perceptual and Motor Skills*, *94*(3), 775–784. https://doi.org/10.2466/PMS.94.3.775-784

Greenland, S., & Brumback, B. (2002). An overview of relations among causal modelling methods. *International Journal of Epidemiology*, *31*(5), 1030–1037. https://doi.org/10.1093/ije/31.5.1030

Greenland, S., Pearl, J., & Robins, J. M. (1999). Causal diagrams for epidemiologic research. *Epidemiology*, *10*(1), 37–48.

Hartman, L. M. (1983). A metacognitive model of social anxiety: Implications for treatment. *Clinical Psychology Review*, *3*(4), 435–456. https://doi.org/10.1016/0272-7358(83)90023-5

Hofmann, S. G. (2007). Cognitive factors that maintain social anxiety disorder: A comprehensive model and its treatment implications. *Cognitive Behaviour Therapy*, *36*(4), 193–209. https://doi.org/10.1080/16506070701421313

Iverach, L, Jones, M., McLellan, L., Lyneham, H., Menzies, R., Onslow, M., & Rapee, R. (2016). Prevalence of anxiety disorders among children who stutter. *Journal of Fluency Disorders*, *49*, 13–28. https://doi.org/10.1016/j.jfludis.2016.07.002

Iverach, L., O'Brian, S., Jones, M., Block, S., Lincoln, M., Harrison, E., Hewat, S., Menzies, R. G., Packman, A., & Onslow, M. (2009). Prevalence of anxiety disorders among adults seeking speech therapy for stuttering. *Journal of Anxiety Disorders*, *23*(7), 928–934. https://doi.org/10.1016/j.janxdis.2009.06.003

Iverach, L., Rapee, R. M., Wong, Q. J., & Lowe, R. (2017). Maintenance of social anxiety in stuttering: A cognitive-behavioral model. *American Journal of Speech-Language Pathology*, *26*(2), 540–556. https://doi.org/10.1044/2016_AJSLP-16-0033

Kefalianos, E., Onslow, M., Ukoumunne, O., Block, S., & Reilly, S. (2014). Stuttering, temperament and anxiety: Data from a community cohort aged 2–4 years. *Journal of Speech, Language, and Hearing Research*, *57*(4), 1314–1322. https://doi.org/10.1044/2014_JSLHR-S-13-0069

Kessler, R. C., Berglund, P., Demler, O., Jin, R., Merikangas, K. R., & Walters, E. E. (2005). Lifetime prevalence and age-of-onset distributions of DSM-IV disorders in the National Comorbidity Survey Replication. *Archives of General Psychiatry*, *62*(6), 593–602. https://doi.org/10.1001/archpsyc.62.6.593

Kikuchi, Y., Kenjo, M., Yoshida, E., Takahashi, S., Murakami, D., Yamaguchi, Y., Adachi, K., Sawatsubashi, M., Taura, M., Nakagawa, T., & Umezaki, T. (2023). Social anxiety disorder in adolescents who stutter: A risk for school refusal. *Pediatrics International*, *65*(1), Article e15622. https://doi.org/10.1111/ped.15622

Kraaimaat, F. W., Vanryckeghem, M., & Van Dam-Baggen, R. (2002). Stuttering and social anxiety. *Journal of Fluency Disorders*, *27*(4), 319–331. https://doi.org/10.1016/S0094-730X(02)00160-2

Lange, T., Vansteelandt, S., & Bekaert, M. (2012). A simple unified approach for estimating natural direct and indirect effects. *American Journal of Epidemiology*, *176*(3), 190-195. https://doi.org/10.1093/aje/kwr525

Langevin, M., Packman, A., & Onslow, M. (2009). Peer responses to stuttering in the preschool setting. *American Journal of Speech-Language Pathology*, *18*(3), 264–276. https://doi.org/10.1044/1058-0360(2009/07-0087)

Langevin, M., Packman, A., & Onslow, M. (2010). Parent perceptions of the impact of stuttering on their preschoolers and themselves. *Journal of Communication Disorders*, *43*(5), 407–423. https://doi.org/10.1016/j.jcomdis.2010.05.003

Lowe, R., Jelčić Jakšić, S., Onslow, M., O'Brian, S., Vanryckeghem, M., Millard, S., Kelman, E., Block, S., Franken, M. -C., Van Eerdenbrugh, S., Menzies, R., Shenker, R., Byrd, C., Bosshardt, H. -G., del Gado, F., & Lim, V. (2021). Contemporary issues with stuttering: The Fourth Croatia Stuttering Symposium. *Journal of Fluency Disorders*, *70*, Article 105844. https://doi.org/10.1016/j.jfludis.2021.105844

Lowe, R., Menzies, R. G., Onslow, M., Packman, S., & O'Brian, S**.** (2021). Speech and anxiety management with persistent stuttering: Current status and essential research. *Journal of Speech, Language, and Hearing Research*, *64*(1), 59–74. https://doi.org/10.1044/2020_JSLHR-20-00144

MacKinnon, D. P., Cheong, J., &. Prirlott, A. G, (2012). Statistical mediation analysis. In Cooper, H. E., Camic, P. M., Long, D. L., Panter, A. T., Rindskopf, D. E., & Sher, K. J. (2012) (Eds). *APA handbook of research methods in psychology, Vol 2: Research designs: Quantitative, qualitative, neuropsychological, and biological* (pp. 313–331). American Psychological Association. https://doi.org/10.1037/13620-018

McAllister, J. (2016). Behavioural, emotional and social development of children who stutter. *Journal of Fluency Disorders*, *50*, 23–32. https://doi.org/10.1016/j.jfludis.2016.09.003

McLean, C. P., Asnaani, A., Litz, B. T., & Hofmann, S. G. (2011). Gender differences in anxiety disorders: prevalence, course of illness, comorbidity and burden of illness. *Journal of Psychiatric Research*, *45*(8), 1027-1035. https://doi.org/10.1016/j.jpsychires.2011.03.006

Moscovitch, D. A. (2009). What is the core fear in social phobia? A new model to facilitate individualized case conceptualization and treatment. *Cognitive and Behavioral Practice*, *16*(2), 123–134. https://doi.org/10.1016/j.cbpra.2008.04.002

Norman, A., Lowe, R., Onslow, M., O'Brian, S., Packman, A., Menzies, R., & Schroeder, L. (2023). Cost of illness and health-related quality of life for stuttering: Two systematic reviews. *Journal of Speech, Language, and Hearing Research*, *66*(11), 4414–4431. https://doi.org/10.1044/2023_JSLHR-23-00072

Packman, A. (2012). Theory and therapy in stuttering: A complex relationship. *Journal of Fluency Disorders*, *37*(4), 225–233. https://doi.org/10.1016/j.jfludis.2012.05.004

Park, V., Onslow, M., Lowe, R., Jones, M., O'Brian, S., Packman, A., Menzies, R., Block, S., Wilson, L., Harrison, E., & Hewat, S. (2021). Psychological characteristics of early stuttering. *International Journal of Speech-Language Pathology*, *23*(6), 662–631. https://doi.org/10.1080/17549507.2021.1912826

Rapee, R. M., & Heimberg, R. G. (1997). A cognitive-behavioral model of anxiety in social phobia. *Behavior Research and Therapy*, *35*(8), 741–756. https://doi.org/10.1016/S0005-7967(97)00022-3

Reilly, S., Onslow, M., Packman, A., Cini, E., Conway, L., Ukoumunne, O. C., Bavin, E. L., Prior, M., Eadie, P., Block, S., Wake, M. (2013). Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics*, *132*(3), 460–467. https://doi.org/10.1542/peds.2012-3067

Rijnhart, J. J., Lamp, S. J., Valente, M. J., MacKinnon, D. P., Twisk, J. W., & Heymans, M. W. (2021). Mediation analysis methods used in observational research: a scoping review and recommendations. *BMC Medical Research Methodology*, *21*, Article 226. https://doi.org/10.1186/s12874-021-01426-3

Rothbart, M. K., Ahadi, S. A., Hershey, K. L., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development*, *72*(5), 1394–1408. https://doi.org/10.1111/1467-8624.00355

Schlenker, B. R., & Leary, M. R. (1982). Social anxiety and self- presentation: A conceptualization and model. *Psychological Bulletin*, *92*(3), 641–669. https://doi.org/10.1037/0033-2909.92.3.641

Sjøstrand, Å., Kefalianos, E., Hofslundsengen, H., Guttormsen, L. S., Kirmess, M., Lervåg, A., & Bottegaard Naess, K. -A. (2021). Non-pharmacological interventions for stuttering in children six years and younger. *Cochrane Database of Systematic Reviews*, *9*, Article CD013489. https://doi.org/10.1002/14651858.CD013489.pub2

Slomian, J., Honvo, G., Emonts, P., Reginster, J. Y., & Bruyère, O. (2019). Consequences of maternal postpartum depression: A systematic review of maternal and infant outcomes. *Women's Health*, *15*, Article 1745506519844044. https://doi.org/10.1177/1745506519844044

Song, Z., Huang, J., Qiao, T., Yan, J., Zhang, X., & Lu, D. (2022). Association between maternal anxiety and children's problem behaviors: A systematic review and meta-analysis. *International Journal of Environmental Research and Public Health*, *19*(17), Article 11106. https://doi.org/10.3390/ijerph191711106

Spence, S. H. (1998). A measure of anxiety symptoms among children. *Behaviour Research and Therapy*, *36*(5), 545–566. https://doi.org/10.1016/S0005-7967(98)00034-5

Spence, S. H., & Rapee, R. M. (2016). The etiology of social anxiety disorder: An evidence-based model. *Behaviour Research and Therapy*, *86*, 50–67. https://doi.org/10.1016/j.brat.2016.06.007

Stein, M. B., Baird, A., & Walker, J. R. (1996). Social phobia in adults with stuttering. *American Journal of Psychiatry*, *153*(2), 278–280. https://doi.org/10.1176/ajp.153.2.278

Tennant, P. W. G., Murray, E. J., Arnold, K. F., Berrie, L., Fox, M. P., Gadd, S. C., Harrison, W. J., Keeble, C., Ranker, L. R., Textor, J., Tomova, G. D., Gilthorpe, M. S., & Ellison, G. T. H. (2021). Use of directed acyclic graphs (DAGs) to identify confounders in applied health research: Review and recommendations. *International Journal of Epidemiology*, *50*(2), 620–632. https://doi.org/10.1093/ije/dyaa213

Tığrak, T. K., Kayıkcı, M. E. K., Kirazlı, M. Ç., & Tığrak, A. (2020). Emotional and behavioural problems of children and adolescents who stutter: Comparison with typically developing peers. *Logopedics Phoniatrics Vocology*, *46*(4), 186–192. https://doi.org/10.1080/14015439.2020.1855472

Tomisato, S., Yada, Y., & Wasano, K. (2022). Relationship between social anxiety and coping profile in adults who stutter. *Journal of Communication Disorders*, *95*, Article 106167. https://doi.org/10.1016/j.jcomdis.2021.106167

Tran, Y., Blumgart, E., & Craig, A. (2011). Subjective distress associated with chronic stuttering. *Journal of Fluency Disorders*, *36*(1), 17–26. https://doi.org/10.1016/j.jfludis.2010.12.003

VanderWeele, T. J. (2016). Mediation analysis: a practitioner's guide. *Annual Review of Public Health*, *37*, 17–32. https://doi.org/10.1146/annurev-publhealth-032315-021402

VanderWeele, T. J., Ding, P. (2017). Sensitivity Analysis in Observational Research: Introducing the E-Value. *Annals of Internal Medicine*, *167*(4), 268-274. https://doi.org/10.7326/M16-26

Vanryckeghem, M., & Brutten, G. J. (2007). *KiddyCat: Communication attitude test for preschool and kindergarten children who stutter*. Plural Publishing

Vanryckeghem, M., & Van Eerdenbrugh, S. (2024). What do adults who stutter think about the nature of stuttering treatment? *Perspectives of the ASHA Special Interest Groups*, *9*(2), 320–330. https://doi.org/10.1044/2023_PERSP-23-00030

565 Weidner, M. E., Louis, K. O. S., Burgess, M. E., & LeMasters, S. N. (2015). Attitudes toward
566 stuttering of nonstuttering preschool and kindergarten children: A comparison using a
567 standard instrument prototype. *Journal of Fluency Disorders*, *44*, 74–87.
568 https://doi.org/10.1016/j.jfludis.2015.03.003
569 Węsierska, K., & Vanryckeghem, M. (2015). A comparison of communicative attitudes
570 among stuttering and nonstuttering Polish preschoolers using the KiddyCAT. *Procedia -*
571 *Social and Behavioral Sciences*, *193*, 278–284.
572 https://doi.org/10.1016/j.sbspro.2015.03.272
573 Yaruss, J. S., Quesal, R. W., & Murphy, B. (2002). National Stuttering Association members'
574 opinions about stuttering treatment. *Journal of Fluency Disorders*, *27*(3), 227–242.
575 https://doi.org/10.1016/S0094-730X(02)00142-0

576 **Figure Captions**

577

578 Figure 1. A mediation model of an exposure *E* causing an outcome *O*, and a mediator *M*.

579

580 Figure 2. An exposure *E* causing an outcome *O*, with a confounder *C* that causes E and also
581 causes *O*.

582

583 Figure 3. *C* is a confounder of the mediator M and the outcome *O*.

584

585 Figure 4. *C* is a confounder of the mediator M the exposure *E*.

586

587 Figure 5. Two ways that the mediator-outcome confounder *C* can itself be influenced by the
588 exposure *E*.

589

590 Figure 6. A Directed Acyclic Graph (DAG) summarising our knowledge and hypotheses
591 about the process of how stuttering may lead to social anxiety early in life through
592 negative peer reactions and negative self-perception.