Identification of Risk Factors for Developmental Stuttering

Laura Brompton & Stephanie Gould

Supervisor: Dr. Deryk Beal Reader:

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ABSTRACT

Previous studies on risk factors for developmental stuttering have looked at the correlation between stuttering and a wide variety of factors. External factors such as socio-economic status, cultural factors, and expectations placed on children have all been considered. Internal factors such as mood and temperament have also been examined. Past studies have provided mixed or contradictory results. More research is needed to determine factors that increase the risk for developmental stuttering.

The purpose of the current study is to establish the rationale for the creation and distribution of a short questionnaire to further explore risk factors for developmental stuttering. A short questionnaire will be distributed to the parents of a group of 1000 children available through the Edmonton cohort of the Canadian Healthy Infant Longitudinal Development (CHILD) Study. The CHILD Study is a Canadian longitudinal birth cohort study interested in the effects of environmental and genetic factors on development. The questionnaire will ask parents if their child has ever showed repetitions, lengthening or hesitations in their speech, indicating that the child is currently or has previously experienced developmental stuttering. Results will be compared to existing data in the Edmonton cohort of the CHILD study to examine any possible predictors of developmental stuttering.

The Edmonton cohort of the CHILD study provides a unique and expansive database to use in analyses. We anticipate the richness of the information in the CHILD database will allow for an in-depth analysis on possible predictive factors for developmental stuttering. In addition, we hope that the results of the questionnaire will add to the prevalence literature on developmental stuttering.

BACKGROUND

Stuttering is defined as an atypically high number and/or length of stoppages that disrupt the forward flow of speech (Wingate, 1964). When stuttering begins during early childhood without evidence of a psychological or physiological trauma, it is termed

'developmental stuttering' and is considered idiopathic (van Borsel, 2001). A vast majority of the population, especially children around the age of 2 years old, experience occasional breakdowns in the continuity with which sounds are linked together during continuous speech (Goldman-Eisler, 1968.) However, there are several specific characteristics of the types of fluency breakdowns (termed disfluencies) that are seen in developmental stuttering (Lavid, 2003.)

The core behaviors of developmental stuttering are: sound and syllable repetitions, prolongations of sounds, and blocks (periods where no sound is produced despite attempts at speech) (Wingate, 1964). There are also secondary behaviors that are attempts to escape or avoid the core behaviors, such as: eye blinks, circumlocutions (avoiding problematic words by using alternative words), and interjections (such as "um") (Wingate, 1964). A characteristic unique to developmental stuttering is that the disfluencies typically occur at the beginning of a sentence (Brown, 1938; Jayaram, 1984; Lavid, 2003). The disfluencies are also more likely to be part-word and monosyllabic word repetitions than the multisyllabic word and phrase repetitions, revisions, and interjections seen in the disfluencies of typical speakers (Yairi, 1981).

Developmental stuttering typically begins between the ages of two and four (Månsson, 2000; Reilly, 2009; Yairi & Ambrose, 2013), accompanying a period of substantial language development. The onset of stuttering may begin suddenly or gradually increase in severity (Yairi, Ambrose & Niermann, 1993), with preschool-aged children who stutter exhibiting three times as many disfluencies as typically fluent peers (Tumanova, 2014). Developmental stuttering is prevalent in approximately one percent of the world's population, affecting all countries and languages (Lavid, 2003).

Risk Factors for Onset

Developmental stuttering is a disorder with an elusive cause; although records of its existence date back more than forty centuries ago (Klingbeil, 1939), an explanation of why it affects some children while sparing others remains a mystery. It is most widely believed to be a disorder of the neural physiology underlying speech processes (see Buchel, 2004 for a review). However, the reasons for the underlying neural physiology differences are debated. It is now widely accepted that developmental stuttering is a multifaceted disorder with many causes (Yairi, 2007; Yairi & Ambrose, 2013; Cavenagh, Costelloe, Davis, & Howell, 2015).

Family History. Research has shown genetics play a role in the development of childhood stuttering (see Kraft and Yairi, 2012, for a recent review), with current research exploring the relationship of certain genes to the onset of stuttering in specific families (Drayna, 1997; Shugart, Mundorff, Kilshaw, Doheny, & Doan, et al., 2004). Yairi and Ambrose (2005) found that 65% of children with developmental stuttering had a positive family history of stuttering, with the percentage increasing to 88% in children deemed persistent stutterers. Although it appears multiple genes are involved and the relationship is complex, a family history of stuttering appears to be a strong risk factor for both developing and maintaining a stutter.

Recently, research by Cavenagh, Costelloe, Davis, & Howell (2015) identified that males with developmental stuttering were significantly more likely than females to have a positive family history of stuttering (76% compared to 53%, respectively.) Further research to explore this correlation could lead to better means of identifying children at risk and more definitive diagnoses.

Temperament and emotional characteristics. Emotional factors, specifically emotional vulnerability (Walden, Frankel, Buhr, Johnson, and Conture, 2012), and temperament characteristics (Anderson, Pellowski, Conture, Kelly, 2003), such as shyness, sensitivity, adaptability and vulnerability, have also been suggested as predispositions for developmental stuttering but with limited research and inconclusive results (Reilly, 2009). Many studies that have looked at parent report of their child's temperament have found parents of children who stutter are more likely to rate their children as less able to adapt to new situations compared to their peers (McDevitt & Carey, 1978), and as more emotionally reactive (Karrass, et al., 2006). However, studies that did not rely on parent report have been mixed: some studies that measured physiological responses associated with temperament characteristics (such as the startle response) have found that children who stutter scored higher on measures of arousal (Guitar, 2003), while others have found no difference compared to non-stuttering peers (Alm & Risberg, 2007; Ellis, Finan, & Ramig, 2008). Researchers have not yet explored temperament differences between recovered and persistent sub-types (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015).

Under the most recent revision of *The American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders Revision V Text Revision* (DSM 5) the diagnosis of 'developmental stuttering' was revised to 'childhood onset fluency disorder'. The American Speech-Language-Hearing Association (ASHA) recommended the term 'developmental' be dropped from the description of the disorder as the "disorder is not developmental in nature, but rather is applicable to individuals whose stuttering has an observed onset during childhood"

("ASHA's Recommended Revisions to the DSM-5", 2012, p.15). However, as the majority of the literature still references 'developmental stuttering' as opposed to 'childhood onset fluency disorder', we have chosen to retain the previous terminology. Along with the change of name, the criteria were updated to reflect the common co-occurrence of anxiety and avoidance of social situations (Cohen, 2014). Several studies have found a significant relationship of anxiety and developmental stuttering in adolescent and adult populations (Craig, 1990; Gabel, Colcord, & Petrosino, 2002). However, studies of children have been mixed (Craig & Hancock, 1996; Davis, Shisca, & Howell, 2007; van der Merwe, Robb, Lewis, & Ormond, 2011); in a recent study by Kefalianos, et al (2014), children with developmental stuttering and controls scored equally on measures of anxiety and measures known to be precursors for anxiety. The requirement of anxiety to receive a diagnosis of childhood onset fluency disorder under the DSM 5 should therefore be viewed cautiously as to its necessity.

Parenting Style. Some studies that have examined the characteristics of parents with children who stutter have found small correlations with perfectionistic and demanding personality traits (Moncur, 1952; Darley, 1955) and have found that these parents use faster speaking rates (Meyers, 1985), ask more questions, interrupt their children more frequently and use longer, more complex utterances, but research has been varied (Goodstein, 1956; Kelly & Conture, 1992) and effect sizes have been small (Zebrowski, 1995). It has been hypothesized that parents who place high levels of pressures on their children, whether intentionally or unintentionally, may contribute to the likelihood of stuttering. Conversely, parents who employ fluency-enhancing techniques may mediate the speech environment and reduce the chances of

disfluent speech (Guitar, Schaefer, Donahue-Killburg, & Bond, 1992; Yaruss, Coleman, & Hammer, 2006.)

Socioeconomic Status. Socioeconomic status (SES) of families and, relatedly, parent's education level have also been researched but with mixed empirical support. Two large scale studies were done on socioeconomic status in the twentieth century. Schindler (1955) found 1/3 of children with stutters were in his three highest SES groups; Morgenstern (1956) found the majority of children with stutters belonged to families in the middle of the SES spectrum. More recent studies have had contradictory results, with some studies finding no relationship of SES or parent education level and developmental stuttering (Keating, Keating, Turrell, & Ozanne, 2001; McKinnon, McLeod, & Reilly 2007), with others finding either rising levels of developmental stuttering with maternal education (Reilly, et al., 2009; Howell, 2010) or decreasing levels of developmental stuttering with increasing maternal education (Boyle, et al., 2011). The opposing directions of recent studies should ignite further interest in researching the correlation of SES and parental education level with developmental stuttering onset as there is likely a dynamic relationship with potential mediating factors at play which may affect therapy options. For instance: if SES influences a parent's decision to report stuttering to a professional, as suggested by Yairi & Ambrose (2013) additional resources may need to be established to ensure children from other families are equally as likely to receive treatment.

Language. Due to the overlapping timeline of rapid language development and the onset of developmental stuttering, the connection between language ability and stuttering has been highly researched. In the past, numerous studies concluded that children who stutter were more likely to exhibit speech and language disorders than children who do not stutter

(see Blood, Ridenour, Qualls, & Hammer, 2003, for a review). More current research has criticized the methodology of these studies and questioned the relationship between language ability and stuttering (Nippold, 2012). The *Demands and Capacities Model* theorizes that stuttering is a result of the demands for fluent speech exceeding the child's language, motor or social/emotional capacities (Starkweather & Gottwald, 1990). This model has been used to explain how both children with above average and below average language abilities are at higher risk for onset of stuttering due to the increased cognitive and motoric demands placed on their system (Starkweather et al., 1990). Research has brought mixed results with some studies indicating that children with below average language skills are at a greater risk for continued stuttering (Arndt & Healey, 2001) and others concluding that the presence of a language impairment does not increase the child's risk of stuttering onset and that children who stutter, like other children, present with the full-range of language abilities (Nippold, 2012). As demonstrated by the discrepancy of findings, the relationship between stuttering and language ability is poorly understood and requires further investigation. Even in cases where researchers came up with congruent conclusions, they frequently presented opposing explanations for their findings (Nippold, 2012)

Persistent Stuttering

Approximately seventy to eighty percent of children afflicted with developmental stuttering recover before adulthood (Kloth, Kraaimaat, Janssen, & Brutten, 1999; Yairi & Ambrose, 1999). If stuttering recovers naturally within eighteen months (with no or minimal treatment) the developmental stuttering is deemed 'transitory' as opposed to 'persistent' (when the developmental stuttering persists for more than three years.) Therefore, stuttering is

considered a disorder with two developmental paths: persistent and recovered (Yairi & Ambrose, 2013). Necessarily then, to be 'at a risk' for stuttering could refer to either a risk for stuttering onset or a risk for stuttering persistence, with many studies supporting the belief that persistent stuttering has additional causal factors (Yairi & Ambrose, 2005; Howell & Davis, 2011.) Age is a factor in recovery; recovery typically occurs in the school years and the older a child is the less likely they will spontaneously recover (Lavid, 2003). Gender is also a factor in recovery: although the sex ratio of boys to girls at onset is small (reported at 1.58:1 in a study by Reilly, et al. (2009) and insignificant in a more recent review by Yairi & Ambrose (2012)), boys are less likely to spontaneously recover, with a sex ratio expanding with age and reaching 4:1 by adulthood (Yairi & Ambrose, 1999; Yairi & Ambrose, 2013). Family history also plays a role in recovery: when a child's family includes members whose stuttering persisted beyond childhood, a natural recovery is less common (Ambrose, Cox, & Yairi, 1997). As mentioned previously, the influence of genetics is stronger in those with persistent stuttering compared with those whose stuttering resolves naturally (Yairi & Ambrose, 2005).

Long-term Consequences of Persistent Stuttering. School age children who stutter are at a greater risk for being bullied by peers (Reilly, 2009) and there are several lifelong consequences of having a developmental stutter, such as: educational and occupational underachievement (Yairi, 1997), psychiatric illnesses, such as depression (Santostefono, 1960) and anxiety (Treon et al., 2006), and impaired communication (Yaruss, 2010). To avoid life-long impacts, it is crucial that early intervention be made accessible as soon as possible. Speech services offered to preschool and early elementary populations of children with mild to moderate stuttering have been shown to be effective; however, those who do not receive

stuttering therapy until after puberty typically make only a partial recovery (Andrews, Guitar, & Howie, 1980).

As most children recover naturally, it is important that information about factors that predict early stuttering and factors that predict a natural recovery be found so that speech services are optimized (Reilly, 2013). Identifying risk factors will help determine children at risk for the development and persistence of stuttering as well as optimize and improve services. It is often difficult to pinpoint the exact age at onset of stuttering; an adult who stutters or the parents of a child who stutter are often asked to recall back several years to the moment of stuttering which leads to inaccuracies. A longitudinal study of children identified soon after onset and followed for several years would be of benefit to learn more about factors correlated with stuttering and persistence. Longitudinal studies, in particular, have proven useful to identifying possible risk factors of developmental stuttering in young populations (Yairi & Ambrose, 2005). By accessing a large cohort of Canadian children, this study aims to address factors that may predict the development of developmental stuttering and factors that may predict a natural recovery.

METHOD

Overview of the CHILD Study

The Canadian Healthy Infant Longitudinal Development (CHILD) study examines the influence of genes and the environment on healthy infant development. Expectant mothers, 18 years of age and older, were recruited from the general population in Vancouver, Edmonton, Toronto, Winnipeg and two other rural Manitoban sites. Women were monitored for the remainder of their pregnancy and in-depth information about their child was collected prior to

and immediately after birth. On-going data continues to be collected via medical tests (e.g., spirometry to assess lung function), biological samples (e.g., blood samples) and questionnaires at set intervals (3 months, 6 months, 1 year, 1.5 years, 2 years, 2.5 years, 3 years, 4 years and 5 years) to assess a wide-variety of factors, related to the child's health. Questionnaires have targeted the mother, father and child to gather information about health, medications, diet and stress and to examine environmental factors such as home environment and socio-economic status. The table below summarizes the information gathered to date.

Mother	**Father**	**Child**	**Environment**
- profile / residence - health - medications / vitamins - diet - stress/psychosocial	- health	- health / medications / diet - clinical assessment	- parenting - socio-economic status - home environment - food packaging and preparation

Of particular relevance to this project are questionnaires related to language, parenting, socio-economic status and stress. A schedule for these questionnaires can be seen below:

	1 yr	**2 yr**	**3yr**	**4 yr**	**5 yr**
Language Development Survey		X			
Child Behavioural Checklist		X	X	X	X
BRIEF-P		X	X	X	X
Parent Child Dysfunction Index	X	X	X	X	X

Mother Stress Questionnaire	X	X	X	X	X
Socioeconomic Status Questionnaire	X	X	X	X	X

Participants

A group of approximately 700 children, available through the Edmonton cohort of the CHILD study, will be invited to participate in our study. Based on the inclusion criteria for the CHILD study, these children, born between October 2010 and October 2012, meet the following criteria: all single births; not resulting from in vitro fertilization, born to mothers over the age of eighteen who read, write and speak English; born after thirty-five weeks gestation; born without major congenital abnormalities or respiratory distress syndrome (RDS) and expected to live near the recruitment area for at least one year. These participants are currently between three to five years of age.

Procedure

Parents from the Edmonton cohort of the CHILD study will be invited to participate in our study which involves filling in a very short online questionnaire about their child and stuttering. In an effort to incur minimal obligation to the families, the questionnaire will include five short questions and will be added to the end of a routine online questionnaire. Parents will be provided with a short explanation of this study, consent will be requested digitally, and parents will be informed that a small number of additional questions have been added. Optimally, these questions will be added to the Child Health Questionnaire, which is sent out repeatedly at set intervals as part of the CHILD study. Our study will line up with the final two data collection points, age four and age five, as all children in the Edmonton cohort have

already turned three. For this minimal time commitment, two to five extra minutes, parents will benefit from having their attention drawn to their child's fluency and be provided with a mechanism for referral to a Speech-Language Pathologist (SLP) if necessary.

Questionnaire

Our questionnaire will define stuttering in concise, parent-friendly terminology and, in order to contribute to a cohesive body of knowledge, will be consistent with Reilly et al.'s (2013) study. Parents will be asked three questions about their child's fluency and two questions related to a familial history of stuttering (see Appendix). Parents will be reminded that stuttering can start gradually or suddenly and can persist or recover naturally without treatment. The first three questions will ask if their child has ever showed repetitions, lengthening or hesitations in their speech, indicating that the child is currently or has previously experienced developmental stuttering. The final two questions will ask about the history of stuttering within the family.

If it is expected that the child is stuttering based on a parent's response to the questionnaire, a follow up phone call from the SLP involved with the study will be made. The SLP will explore the nature of the disfluencies further and will make a referral to a community SLP, unless it is clear that the speech behaviours described are not consistent with developmental stuttering or the child has clearly recovered naturally (Reilly et al., 2013). Prevalence and incidence estimates will be made based on the results of the questionnaire and data will be compared to existing data in the Edmonton cohort of the CHILD study to examine any possible predictors of developmental stuttering.

RESULTS AND DISCUSSION

Prevalence and Incidence

Based on the questionnaire, prevalence and incidence figures will be estimated. Determining the prevalence and incidence of stuttering is potentially challenging. Incidence figures differ based on the time period observed and when in the lifespan the observation takes place (Yairi & Ambrose, 2013). During the preschool years when onset is most common, incidence (new cases within a given time period) may exceed prevalence (those currently stuttering) as children who stuttered in the past have now recovered, leaving less current stutterers. The opposite tends to be true in the teenage years when new onsets are minimal and stuttering tends to persist (Yairi & Ambrose, 2013).

Incidence figures are difficult to compare as some may report incidence for various time periods while others present figures relating to the entire lifespan. This variation is evident, with recent (since 2000) literature reporting preschool incidence rates as low as 2.8% (Craig, et. al., 2002) and ranging to as high as 17.7% (Yairi & Ambrose, 2013). It appears that incidence rates are highest in studies which occur closest to age of onset and in studies with rigorous evaluation processes. In the past, five percent incidence seems to be the most generally agreed upon and accepted statistic but recent studies indicate that figure as too conservative with new incidence figures centering around eight percent (Yairi & Ambrose, 2013). Reilly's large-scale, Australian-based study provides a recent incidence estimate of children who stutter before age three to be 8.5% (Reilly, 2009). This figure was higher than previous reports which the authors attributed to the prospective design of the longitudinal study (Reilly, 2013). Based on these previous studies we predict our questionnaire will yield a figure which is comparable to Reilly's 8.5%.

Prevalence figures are easier to obtain but still fluctuate based on a number of factors including the age of the sample, the definition of stuttering and the method of identifying stuttering (Yairi & Ambrose, 2013). Recent prevalence estimates confirm prevalence is highest in the preschool age group with figures falling between two and three percent. Prevalence later in life is much lower, due to recovery and seems to hover around the one percent mark. Only one life-span prevalence figure is available at 0.72% (Yairi & Ambrose, 2013). It is expected that this study will yield a prevalence estimate of between two and three percent.

Prevalence and incidence measures are further affected by the definition of stuttering that is used. There has been much debate in the field of stuttering about the inclusion of whole-word repetitions in the definition of stuttering. Howell (2010) criticized Reilly et al. (2009) for the inclusion of whole-word repetitions in her definition of stuttering. Inclusion and exclusion criteria of the study also influence these measures. For instance, we know congenital factors, such as physical trauma in utero or at birth, Cerebral Palsy, Down's syndrome or childhood surgery, have been linked to a greater risk of developmental stuttering (Poulos & Webster, 1991), however; these children are commonly omitted from stuttering research studies. The exclusion criteria of the CHILD study omits these children and therefore we would predict slightly fewer cases of stuttering to be reported. We predict this decrease to be insignificant. This study will add uniquely Canadian data to the body of evidence on stuttering and allow prevalence and incidence figures to be compared to those in other parts of the world, such as Australia.

Correlation of Stuttering and Other Factors

Age. Along with prevalence and incidence data, we believe our study will add to the body of literature on the correlation of stuttering and other factors. The most robust correlate for developmental stuttering is age, as illustrated by the prevalence and incidence figures reported above. We expect the results from our questionnaire will support the evidence that stuttering onset most commonly occurs between the ages of two and four (Månsson, 2000; Buck, Lees, & Cook, 2002; Yairi and Ambrose, 2005; Reilly, et al., 2009).

Family History. Several areas of research, including family studies, twin studies and molecular research, have demonstrated strong evidence for the genetic predisposition for stuttering (see Yairi and Ambrose, 2012). Although the underlying genetic mechanisms are not fully understood, it is widely accepted that both onset and persistence of stuttering are influenced by genetics. As a, a family history of stuttering is expected in many of the cases where stuttering-like disfluencies are noted by parents. In order to examine the contribution of heritability, it is important that our questionnaire inquires about a family history of developmental stuttering. The CHILD study has collected a wealth of information about the parents who are involved with this study. They have not, however, asked specific questions regarding a family history of developmental stuttering. Based on previous literature, we expect that the incidence of stuttering in first-degree relatives, parents or siblings, will be approximately fifty percent (Buck, Lees, & Cook, 2002; Riaz et al., 2005; Yairi & Ambrose, 2005) increasing to sixty or seventy percent when more distant relatives are included (Månsson, 2000; Yairi & Ambrose, 2005). Due to the age of the children involved in this study, it is not possible to say with certainty who is a persistent stutterer. Children, who are currently stuttering at the time of this study, may go on to stutter indefinitely or may recover

spontaneously or with the help of speech-language therapy. Children who are reported to have stuttered in the past but no longer stutter at the time of the study will be considered recovered stutterers; however, it is important to consider the reliability of these cases as they are based solely on retrospective parent report. Regardless of the challenges of categorizing these children as persistent or non-persistent stutterers, it is expected that the children who go on to persist in their stuttering have an even stronger genetic influence. Yairi and Ambrose (2005) report 88% of persistent stutterers having a positive family history of stuttering.

Gender. Consistently throughout history, a significant gender bias for stuttering has been reported in the literature - with male-to-female ratios of 4:1 or higher (Yairi & Ambrose, 1999; Yairi & Ambrose, 2013). Upon closer examination, however, it appears that gender ratios are very small and perhaps even insignificant near the age of onset (see Yairi & Ambrose, 2012). Yairi & Ambrose reviewed five studies involving preschool-aged children who stutter and found the difference between numbers of males versus females to be statistically insignificant. Based on this finding, we expect our questionnaire to uncover comparable numbers of boys and girls who stutter. Gender bias increases considerably with age (Yairi & Ambrose, 1999; Yairi & Ambrose, 2013). High male-to-female ratios in adolescents and adults who stutter indicate that females who stutter are more likely to recover than males. Females are also reported to recover more quickly than males resulting in females who stutter for more than one year to be at higher risk for persistence (Yairi & Ambrose, 1999).

Recently, Cavenagh, Costelloe, Davis, & Howell (2015) found a higher percentage of positive family history in males with developmental stuttering as compared to females. We are

optimistic that our study will find similar results and will add to this newly discovered correlation.

Factors Receiving Mixed Support

The correlation of developmental stuttering with age, family-history and gender is well documented in the literature. We expect our Canadian data will align with the previous literature and contribute to the evidence for these universal correlates. Of great interest is the relationship between factors that have been hypothesised in the literature but have received mixed support. The information already obtained in the CHILD cohort will allow us to examine factor such as mother's reported stress levels and psychosocial health, parenting attitudes, socio-economic status, parents' level of education, child's reported temperament and personality characteristics, and the child's language development and health history, all of which have been studied but with varied conclusions.

Parenting Styles/Stress. Leftover thinking from the diagnosogenic theory of stuttering, suggests that level of pressure exerted on children significantly impacts their probability of stuttering (Johnson, 1956; Bloodstein, 1995). In the past, various studies have attributed children's stuttering to different sources of stress including demanding or critical parenting and cultural pressure to perform (Moncur, 1952; Darley, 1955; Johnson, 1956). The results from these studies have been mixed and sometimes conflicting but seem to point to the role of stress in developmental stuttering. While current stuttering theory does not agree with the diagnosogenic premise that stuttering is a result of parent's overreaction to their children's normal disfluencies, it is possible that external stressors placed on the child contributes to the expression of stuttering in a child who already possesses the genetic propensity to stutter (Lee,

2014). Historical studies have been criticized for being influenced by researcher expectations (Finn & Cordes, 1997). The CHILD study provides an unbiased record of parental stress levels during pregnancy and the first five years of participant's lives, thus providing a unique opportunity to compare objective measures of stress levels and onset of developmental stuttering. The rich environmental information collected about our participants may also provide clues as to environmental or cultural factors which influence onset of stuttering in the preschool years.

Temperament and Emotional Characteristics. Research on temperament that has used parent report has shown higher ratings on measures related to 'difficult' temperament characteristics, such as lower abilities to adapt to novel situations (McDevitt & Carey, 1978) and higher emotional reactiveness (Karrass, et al., 2006). Research that has not relied on parent report has had more mixed results, with some studies finding no difference in temperament between children who stutter and controls (Alm & Risberg, 2007; Ellis, Finan, & Ramig, 2008). As the CHILD study is based primarily on parent report, we expect to find a correlation between 'difficult' temperament (such as higher sensitivity, restlessness, and impulsiveness) and childhood stuttering.

Health Factors: Sleep and Asthma. In a recent review of factors that may be linked to developmental stuttering onset, Cavenagh, et al. (2015) reported that health history has been under documented in past research. The authors mention that of the limited past research, asthma and sleep problems are factors that have been researched but no significant correlations have been found. Specifically, Rustin and Purser (1991) found eleven percent of children with developmental stuttering enrolled in a clinic had asthma, one third of the boys

had sleep problems, and one fifth of the girls had sleep problems; findings from the general population are equivalent. The exclusion criteria of the CHILD study would eliminate children with serious birth or medical issues; the CHILD study has, however, taken extensive data about sleep and asthma from participants. Based on the study by Rustin and Purser (1991), we expect no correlation of sleep and asthma with developmental stuttering.

Co-occurrence with Disordered Language. While some studies have found that children who stutter score lower than peers on language measure such as: age at first words, age at first sentences, size of receptive vocabulary, mean length of utterance and expressive and receptive syntax (Andrews & Harris, 1964; Arndt & Healey, 2001; Anderson & Conture, 2000), other studies have found no language differences (Nippold, 2012). Bernstein & Ratner (1997) and Bloodstein & Ratner (2008) concluded that differences, which have been demonstrated between groups, have been subtle. Regardless of how they compare to children who do not stutter, it has been demonstrated that as a group children who stutter still score within normal limits on tests of expressive language (Rommel, Häge, Kalehne, & Johannsen, 2000; Watkins, Yairi, & Ambrose, 1999). The CHILD study examines language through the *Language Development Survey* (LDS) which is conducted at 24 months (Rescorla, 1989). This questionnaire uses parent report of vocabulary and word combinations to gather information about the child's language development. The LDS provides parents with a list of 310 words grouped in fourteen different semantic categories (Rescorla & Alley, 2001). The parent decides which of these words are used by their child. While LDS has proven to be a reliable, valid and effective method of identifying language delay in children aged three and under, (Rescorla & Alley, 2001) it is general screening tool which is unlikely to pick up the minor differences in

language between children who stutter and children with normal fluency. It is likely that a more comprehensive speech and language standardized tests would need to be administered to uncover the subtle differences in language and speech development of children who stutter and their typically developing peers. Based on the complex interaction between language ability and stuttering and the nature of the LDS, we do not expect to see any differences in speech and language measures between the two groups.

Past studies that have examined the relationship between language ability and stuttering have been criticized for failing to match participants based on parent's education level (Nippold, 2012). Not uncommonly, the control group was recruited from university neighbourhoods with high socioeconomic status and the experimental group was recruited from community clinics which confound the conclusions that were made about discrepancies in language abilities between the two groups (Nippold, 2012). The CHILD project with its large, diverse population of participants, who were recruited using unbiased methods, provides a unique opportunity to compare the language ability of children who stutter with children who do not stutter without the aforementioned confounding variables. We predict our findings will align with Nippold's (2012) conclusion that children who stutter present with a full range of language abilities and that children who stutter are not more likely to present with a co-occurring language disorder.

Socioeconomic Status and Parents' Level of Education. Early research on developmental stuttering and socioeconomic status/parents' level of education showed a trend for an increasing risk of developmental stuttering with rising SES (Schindler, 1955; Morgenstern, 1956). More recent research has been mixed: studies have found the same trend as earlier

studies (Reilly, et al., 2009; Howell, 2010), an opposite trend (Boyle, et al., 2011), or no correlation (Keating, Keating, Turrell, & Ozanne, 2001; McKinnon, McLeod, & Reilly 2007). When socioeconomic status is separated from parental education level, it has been found to be a weaker predictor of developmental stuttering (Richels et al., 2013.) However, parental education level has been noted to be at risk for a response bias, with parents with higher education levels more likely to report their child's disfluencies (Morgenstern, 1956). The format of the CHILD study should minimize the chances of a response bias. We expect any correlation we find between SES or parental education levels and stuttering will be better explained by increased stress levels in the homes (i.e., homes with higher stress levels will have higher incidence of stuttering onset.)

DISCUSSION

Strengths

This study's strength lies in the wealth of information which has been collected about its participants. The ability to compare children who do and don't stutter to a multitude of aspects of health and development provides a unique opportunity to uncover new predictive factors for onset of developmental stuttering. Current theories of stuttering suggest that onset of developmental stuttering is a result of genetically-mediated neuroanatomical difference and developmental and environmental factors. The CHILD project is designed to examine the relationship between conditions within a child's home and health at five years of age. Because of this, the study has collect rigorous data about the environment in which these preschool-aged children spend their lives. This database of information about each child involved in the

study provides the opportunity to uncover correlations between environmental factors and stuttering.

Limitations

This study has some limitations due it being an add-on to an existing longitudinal study. Participant recruitment was part of the CHILD project and therefore inclusion / exclusion criteria were previously established. Overall the CHILD project has experienced a 5.7 % attrition rate, composed of approximately three percent being excluded based on inclusion criteria and an additional three percent being true attrition from participants leaving the study. In addition, the CHILD project is designed to follow children until age five. Beginning in October of 2015, participants reached five years of age, meaning all assessments and questionnaires are complete for those children. The timing of this add-on study means we will have access to a large number of participants but a small number of the oldest participants will not be available to answer our questionnaire.

Another limitation is the retrospective nature of this study. Onset of developmental stuttering typically occurs between two and six years of age. The participants are now between the ages of three and five meaning it is possible that we may have missed the onset of some participants stuttering. Parent report has proven to be a reliable method of data collection with the most accurate results being given when the behaviour is present (Dale, Bates, Reznick, & Morisset, 1989). Retrospective data has been shown to be less reliable but is easier and more cost-effective to obtain. For this study, parents will be asked to reflect back on their child's fluency and it is possible that some cases of naturally recovered stuttering will be forgotten and therefore not reported. It has been suggested that the most accurate calculations of stuttering

come from prospective studies, starting when children are twelve months of age, in which parents are instructed to look for symptoms of stuttering as they arise (Yairi & Ambrose, 2013). This being said, the parents involved with the CHILD study are accustomed to reporting on various aspects of their child's health and may have more thorough records of their child's behaviour than other parents. Based on this and the fact that the participants are still preschool-aged, it is expected that only a very small number of cases of stuttering will be missed from being reported.

CONCLUSION

Access to a large cohort of Canadian, preschool-aged children will present a unique opportunity to add a Canadian statistic to the prevalence and incidence literature. We predict these figures will be comparable to those from other countries around the world. In addition, the wealth of information available about these children from before birth to five years of age provides considerable data which can be analyzed to predict factors which are correlated with onset of developmental stuttering. We predict our findings will align with well-established risk factors for developmental stuttering such as: onset occurring most commonly between ages two and four, family history of stuttering and rates of spontaneous recovery being higher in girls than boys. It is also expected that this study will contribute new information about the correlation of stuttering with other less agreed upon risk factors such as: parental stress levels, child's sleep patterns, asthma and temperament. These factors have brought mixed results in the literature and warrant further examination to determine their relationship to developmental stuttering. Learning more about these relationships could contribute to a body of existing knowledge about risk factors for developmental stuttering. Increased knowledge of

risk factors is an important contribution to what we currently know about developmental stuttering in that it serves to influence the practice of speech-language pathology. Understanding of potential risk factors contributes to an accurate and confident diagnosis of developmental stuttering and also helps to distinguish children who are likely to recover naturally from those who are likely to persist.

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APPENDIX: QUESTIONNAIRE PROVIDED TO PARENTS ABOUT THE ONSET OF STUTTERING

Children who stutter have trouble getting their words out. Stuttering can start suddenly or gradually (over days, weeks or months) and can persist or recover naturally without treatment. Think about how your child speaks now and how they have in the past.

1. Does your child / has your child ever repeated words or parts of words (e.g. “I - I - I - I - I want to go now.”)?
2. Does your child / has your child ever lengthened sounds within a word (e.g. “I waaaaaaaaaant to go now.”)?
3. Does your child / has your child ever had “blocks” where no sounds come out?
4. Based on the definition above, do you believe that any member of your family has ever stuttered? Please indicate the relationship of this family member to your child.
5. If a member of your family stuttered in the past, did their stuttering persist? (i.e. - Do they still stutter now?)

If your answers indicate your child may stutter or may have stuttered in the past, an SLP affiliated with the CHILD study will contact you. A follow up phone call will take place and a plan for further services (if needed) will be made.