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Young children's family history of stuttering and their articulation, language and attentional abilities: An exploratory study

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Abstract

Purpose—The purpose of this study was to determine whether young children who do (CWS) and do not stutter (CWNS) with a positive versus negative family history of stuttering differ in articulation, language and attentional abilities and family histories of articulation, language and attention related disorders.

Method—Participants were 25 young CWS and 50 young CWNS. All 75 participants' caregivers consistently reported a positive or negative family history of stuttering across three consecutive time points that were about 8 months apart for a total of approximately 16 months. Each participant's family history focused on the same, relatively limited number of generations (i.e., participants' parents & siblings). Children's family history of stuttering as well as articulation, language, and attention related disorders was obtained from one or two caregivers during an extensive interview. Children's speech and language abilities were measured using four standardized articulation and language tests and their attentional abilities were measured using caregiver reports of temperament.

Results—Findings indicated that (1) most caregivers (81.5% or 75 out 92) were consistent in their reporting of positive or negative history of stuttering; (2) CWNS with a positive family history of stuttering, compared to those with a negative family history of stuttering, were more

likely to have reported a positive family history of attention deficit/hyperactivity disorder (ADHD), and (3) CWNS with a positive family history of stuttering had lower language scores than those with a negative family history of stuttering. However, there were no such significant differences in family histories of ADHD and language scores for CWS with a positive versus negative family history of stuttering. In addition, although 24% of CWS versus 12% of CWNS's caregivers reported a positive family history of stuttering, inferential analyses indicated no significant differences between CWS and CWNS in relative proportions of family histories of stuttering.

Conclusions—Finding that a relatively high proportion (i.e., 81.5%) of caregivers *consistently* reported a positive or negative family history of stuttering across three consecutive time points should provide some degree of assurance to those who collect such caregiver reports. Based on such consistent caregiver reports, linguistic as well as attentional vulnerabilities appear associated with a positive family history of stuttering, a finding that must await further empirical study for confirmation or refutation.

Since the mid-1960's, there has been a growing body of empirical evidence indicating that there is familial and/or genetic transmission of vulnerability to stuttering (e.g., Ambrose, Cox, & Yairi, 1997; Andrews & Harris, 1964; Buck, Lees, & Cook, 2002; Cox, Kramer, Kidd, & Rao, 1984; Kraft & Yairi, 2011). Initially, researchers studied such possible genetic contributions to stuttering by means of family history (e.g., Ambrose et al., 1997; Buck et al., 2002) as well as twin studies (e.g., Felsenfeld et al., 2000; Howie, 1981a, 1981b). In a review of 23 studies using the family history method published between 1924 and 1983, Yairi, Ambrose and Cox (1996) concluded that familial stuttering was apparent in 30% to 60% of people who stutter (PWS; range = 20% – 74%) as compared with less than 10% of people who do not stutter (PWNS; range = 1.3% – 42%). It may be challenging to determine accurate percentages of familial stuttering among PWS versus PWNS given the methodological differences across studies, which may have contributed to the disparate findings. For instance, in our informal assessment of 21 accessible studies of a family history of stuttering (published between 1937 and 2011; see Table 1), between-study differences seem to be associated with: (1) participants' chronicity status (i.e., persistent versus recovered stuttering), (2) the extent of family history data collection (i.e., immediate versus extended family members), and (3) varying data informants (i.e., whether family history was collected from caregivers versus the participant). Overall, based on these studies, it appears that a positive family history of stuttering was more apparent (i.e., higher percentage) in studies that (1) included participants with persistent stuttering, (2) gathered information about extended family members, and/or (3) were based on caregivers' reports. It should be noted that more formal, systematic analyses are needed to confirm this informal assessment. Using the twin study method, Howie (1981a) reported that the estimated risk of stuttering in identical twins was higher (.77) than that in fraternal twins (.32). Likewise, Felsenfeld et al. (2000) reported that approximately 70% of the variance in liability to stuttering was accounted for by additive genetic effects, with the remainder by non-shared environmental effects. More recently, researchers have employed other techniques such as biological genetics. For example, systematic analysis of genetic variation indicated that there are 10 significant candidate genes associated with persistent developmental stuttering (Kraft & Yairi, 2011). Thus, whether findings are based on family histories, twin studies or more

modern-day genetic investigations, genetic processes appear to be associated with childhood stuttering.

Nature of Vulnerability to Stuttering

As shown above, converging lines of research suggests a genetic predisposition toward stuttering. However, what remains unclear is the “*nature*” of the vulnerability that prompts stuttering and is transmitted genetically (Yairi & Ambrose, 2005, p.302). Yairi and Seery (2015) proposed several possible vulnerabilities including structural and functional brain features, speech motor control, as well as personality or temperamental characteristics. Based on parent reports of a family history of stuttering, Subramanian and Yairi (2006) noted that stuttering and high-risk (i.e., individuals who do not stutter with a positive family history of stuttering) groups used different speech motor control strategies than those who do not stutter with a negative family history of stuttering. In contrast, Janssen, Kraaimaat and Brutten (1990) found no significant difference in reading abilities (e.g., errors and comprehension) or anxiety levels exhibited by school-age children who stuttered with a positive versus those with a negative family history of stuttering. Similarly, Seider, Gladstien, and Kidd (1982) reported no significant difference in the frequency of language problems between adults who stutter with a positive versus those with a negative family history of stuttering.

The above studies assessed the nature of stuttering vulnerabilities among school-age children and adults who stutter with a positive versus negative family history of stuttering. To date, however, relatively few have examined the nature of vulnerabilities among young children with a positive versus negative family history of stuttering. It is possible that some vulnerabilities (e.g., linguistic or attentional vulnerabilities) are more apparent during early childhood and diminish with maturation. This means that some vulnerabilities that may have triggered the onset of stuttering in young children may not necessarily be observed at a later age. Therefore, young children’s vulnerabilities should be investigated independently of those of older children or adults.

The purpose of this study was to better understand the nature of stuttering vulnerabilities, which may be familial. As a first step toward understanding such familial vulnerabilities, the present study investigated young children with and those without family histories of stuttering, relative to the following variables that have been found to be associated with childhood stuttering: 1) articulation (e.g., Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Blood, Ridenour, Qualls, & Hammer, 2003), 2) language (e.g., Ambrose et al., 2015; Ntourou, Conture & Lipsey, 2011) and 3) attentional processes (e.g., Clark, Conture, Walden, & Lambert, 2015; Felsenfeld, van Beijsterveldt, & Boomsma, 2010).

Such vulnerabilities and their impact on stuttering are accounted for by the relatively recent model of childhood stuttering: the *Dual Diathesis-Stress Model of Childhood Stuttering* (DD-S model, for overview of the model see: Conture & Walden, 2012; for experimental tests of the model, see: Choi, Conture, Walden, Jones & Kim, 2016; Walden, Frankel, Buhr, Johnson, Conture & Karrass, 2012). Briefly, the DD-S model proposes that children’s inherent speech-language and temperamental vulnerabilities interact with external linguistic

and emotional stress respectively, thus contributing to the onset and development of childhood stuttering. For example, the DD-S model predicts that children with lower language abilities would show greater increase in stuttering frequencies when placed under greater linguistic demands compared to children with higher language abilities. However, such differential effects of linguistic demands depending on the child's language abilities would be less likely to be observed when linguistic demands are low.

Overall findings across several empirical studies/reviews suggest that, despite equivocal findings, childhood stuttering might be associated with delayed speech-language abilities (e.g., Pellowski, Anderson & Conture, 2000; Bloodstein & Ratner, 2008; Murray & Reed, 1977; Ntouriou et al., 2011; Ratner & Silverman, 2000) and poorer attentional processes (e.g., Eggers, Luc, Van den Bergh, 2010, 2012; Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Karrass et al. 2006; Jones et al., in press). For example, Arndt and Healey's (2001) survey results from 241 speech-language pathologists revealed that 14.1% of children who stutter (CWS) have phonological disorders (compared to 2–25% of children who do not stutter [CWNS], Law, Boyle, Harris, Harkness, & Nye, 2000), 15.4% of CWS have language disorders (compared to 2.63% to 16% of CWNS, Law et al., 2000) and 14.3% of CWS have both phonological and language disorders. Additionally, about 4% (Arndt & Healey, 2001) to 26% (Riley & Riley, 2000) of CWS are reported to have attention-deficit/hyperactivity disorders (ADHD) whereas 2.7% of CWNS have ADHD (Paster, Reuben, Duran, & Hawkins, 2015). Perhaps, these developmental processes (e.g., language & attention), whether acting alone or in combination contribute to some children's predisposition toward stuttering and/or are associated with a positive family history of stuttering.

Interestingly, studies have shown that speech-language or attentional vulnerabilities are likely to be transmitted in individuals with a positive family history of related disorders. (Lewis et al., 2006; Thapar, Holmes, Poulton, & Harrington, 1999). For example, Felsenfeld, McGue and Broen (1995) reported that 42% of children whose parent had an articulation disorder as a child exhibited "low-average" or "poor" performance on an articulation test whereas only 19% of the control children showed such low performance. Similarly, Alaraifi, Kamal, QA'Dan, & Haj-Tas (2014) found that 46.7% of patients (ages 6 to 35 years old) with articulation disorders reported a family history of functional articulation disorders. Regarding family histories of language disorders, Tallal, Townsend, Curtiss, and Wulfeck (1991) reported that approximately 65% (42/65) of the language-impaired children had a positive family history of language disorders; however, there were no data from non-language impaired children reported in this study. Regarding family histories of ADHD, two retrospective studies reported that 20% to 21% of hyperactive children had a parent who was hyperactive as a child whereas 2% to 5% of controls had parents with the same symptoms (Cantwell, 1972; Morrison & Stewart, 1971). In addition, some studies have shown cross-disorder familial risk. For example, Neils and Aram (1986) reported that compared to the control group, children with *language disorders* had significantly more caregiver reports of the presence of family histories of *stuttering* as well as speech, reading and language disorders.

Purpose of the study

Therefore, the purpose of the present study was to determine *between-group* (CWS vs. CWNS) differences in family histories for: (1) stuttering; and (2) articulation, language and attention disorders. This study also attempted to determine *within-group* differences (i.e., separate, within talker group analyses) for children with a positive versus negative family history of stuttering in: (3) family histories of articulation, language and attention disorders and (4) articulation, language, and attentional abilities.

To address the aims of this study, we investigated the following six research questions:

Between-group comparisons (CWS vs. CWNS):

Research question 1 (CWS vs. CWNS): Do CWS, when compared to CWNS peers, significantly differ in *family histories of stuttering*?

Research question 2 (CWS vs. CWNS): Do CWS, when compared to CWNS peers, significantly differ in *family histories of disorders of (1) articulation, (2) language, or (3) attention*?

Within-group comparisons (Within CWS or CWNS):

Research question 3 (CWS with a positive vs. negative family history of stuttering): Do CWS with a positive family history of stuttering, significantly differ in *family histories of disorders of (1) articulation, (2) language, or (3) attention*, compared to CWS with a negative family history?

Research question 4 (CWNS with a positive vs. negative family history of stuttering): Do CWNS with a positive family history of stuttering, significantly differ in *family histories of disorders of (1) articulation, (2) language, or (3) attention*, compared to CWNS with a negative family history?

Research question 5 (CWS with a positive vs. negative family history of stuttering): Do CWS with a positive family history of stuttering, significantly differ in their (1) articulation, (2) language, and (3) attentional *abilities*, compared to CWS with a negative family history?

Research question 6 (CWNS with a positive vs. negative family history of stuttering): Do CWNS with a positive family history of stuttering, significantly differ in their (1) articulation, (2) language, and (3) attentional *abilities*, compared to CWNS with a negative family history?

The present study used caregiver reports to obtain family history information. Although findings from verbal reports of a family history have yielded results consistent with those obtained from more objective methods (e.g., DNA/genetic evidence), the reliability and validity of caregivers' reports have been questioned by some (e.g., Kraft & Yairi, 2011; Yairi et al., 1996) because: (1) the information is usually not verified by other family members; (2) it may result in false-negative or false-positive identification (e.g., Hedges et al., 1995) and (3) results may be affected by additional confounding variables such as family size. Thus, Ambrose et al. (1997) suggested using more rigorous verification procedures to

increase the accuracy of such data, which involve collecting data from multiple informants and interviews across time.

The present study addressed the above concerns by only including the following family history reports: (1) caregiver reports that *consistently* indicated a positive or negative family history of stuttering across three successive time points, 8 months apart, spanning a total 16 months; and (2) caregiver reports addressing the stuttering history of immediate or first-degree relatives (parents & siblings). Despite these more rigorous methods, we do not contend that family history interviews yield identical data as those obtained from biological/genetic studies.

Method

Participants

Participants were 75 monolingual, English-speaking children (46 boys, 29 girls; 25 CWS, 50 CWNS) aged 36 to 71 months at the time of initial testing. Among them, 61 participants were Caucasian, 7 were African American, 2 were Asian, and 5 were multiracial. Study inclusion criteria, group classification, recruitment strategy, and participant selection procedure are described below.

Study Inclusion Criteria

To prevent confounding factors from affecting the results and to increase the reliability of caregiver-reported family history data, we employed the following inclusion criteria.

Criteria for consistent reports of family history—The following two consistency criteria were employed to minimize the frequency of false positive or false negative identification of family histories of stuttering or articulation, language, and attention-related disorders: (1) Participants included in the study's analyses completed three diagnostic visits, and (2) their caregivers *consistently* reported either the presence or absence of a family history of stuttering among first-degree relatives (i.e., immediate family including parents, and siblings). We included family history information from immediate family members only, to minimize the variation of family sizes across families (Kraft & Yairi, 2011) and to prevent possible inaccuracies associated with recall of stuttering among extended family members if those individuals are not present to verify such information (Ambrose et al., 1997). Reports were considered consistent if they were identical across the three time points about 8 months apart over the course of approximately 16 months. For research questions involving family histories of articulation, language and attention-related disorders (i.e., research questions 2, 3, & 4), children with inconsistent reports of family histories of those disorders were additionally excluded.

Criteria for articulation, language, and hearing abilities—The following articulation, language and hearing criteria were employed to prevent confounding factors from affecting the results. Participants performed within normal limits (i.e., scored at the 16th percentile or higher) on the (1) Peabody Picture Vocabulary Test – Fourth Edition (PPVT; Dunn & Dunn, 2007), (2) Expressive Vocabulary Test – Second Edition (EVT;

Williams, 2007), (3) Test of Early Language Development – Third Edition (TELD; Hresko, Reid, & Hamill, 1999), and (4) Sounds in Words subtest of the Goldman-Fristoe Test of Articulation 2 (GFTA; Goldman & Fristoe, 2000). Furthermore, each child passed a bilateral pure tone hearing screening. Fisher's Exact Test indicated no significant between-group (CWS vs. CWNS) differences in the rates of children with below-normal-limit articulation (based on GFTA standard scores, $p = 1.00$, Cramer's $V = .002$) or language abilities (based on composite language scores, see p.14 for calculation of composite language scores; $p = .772$, Cramer's $V = .031$).

By ensuring that all participants performed within normal limits, the authors mitigate the possibility that a chance disproportionate representation of articulation or language disorders in any of the participant subgroups could in turn confound the results. Such a disproportionate representation would be particularly problematic when analyzing the family histories of articulation and language disorders in CWS versus CWNS. This is due to the possibility that children with below-normal-limit articulation or language abilities are more likely to have positive family histories of articulation or language disorders respectively (Felsenfeld et al., 1995; Tallal et al., 1991).

Criteria for developmental and treatment history—No participant had received any known or reported formal treatment for stuttering or other communication disorders prior to participation. Likewise, no participant had any known or reported neurological, developmental, academic, intellectual, or emotional problems.

Group Classification

Groups based on stuttering diagnosis: CWS versus CWNS—Participants were considered CWS if they (a) exhibited three or more stuttered disfluencies during conversational speech based on a 300-word sample (Conture, 2001) *and* (b) received a total overall stuttering severity score of 11 or above (i.e., a severity equivalent of at least “mild”) on the Stuttering Severity Instrument-3 (SSI-3, Riley, 1994). Participants were considered CWNS if they (a) exhibited two or fewer stuttered disfluencies during conversational speech based on a 300-word sample, *and* (b) received a total overall stuttering severity score of 10 or below (i.e., a severity equivalent of “very mild”) on the SSI-3.

Groups based on family history of stuttering—Participants were considered to have a positive family history of stuttering if their caregivers consistently reported a *presence* of a family history of stuttering among first-degree relatives (i.e., parents & siblings) once every 8 months for three time points. Participants were considered to have a negative family history of stuttering if their caregivers consistently reported an *absence* of a family history of stuttering among first-degree relatives (i.e., parents & siblings) once every 8 months for three time points.

Recruitment Strategy

Participants were recruited through the following methods in the metropolitan Nashville, Tennessee area: (a) a free, regionally-distributed parent-oriented magazine, (b) local health care provider referrals, or (c) self/professional referral to the Vanderbilt Developmental

Stuttering Laboratory. Participants were part of an ongoing series of investigations of linguistic and emotional contributors to developmental stuttering conducted by the Vanderbilt University Developmental Stuttering Project (Arnold, Conture, Key, & Walden, 2011; Choi, Conture, Walden, Lambert, & Tumanova, 2013; Choi et al., 2016; Clark, Conture, Frankel & Walden, 2012; Clark, Conture, Walden, & Lambert, 2013, 2015; Johnson, Walden, Conture, & Karrass, 2010; Jones et al., 2014; Jones, Conture, & Walden, 2014; Jones et al., in press; Ntourou, Conture, & Walden, 2013; Tumanova, Conture, Lambert & Walden, 2014; Zengin-Bolatkale, Conture, & Walden, 2015). The study's protocol was approved by the Institutional Review Board of Vanderbilt University. Caregivers signed informed consent and children gave assent for participation.

Participant selection procedure

From an initial pool of 195 participants (15 CWS females, 64 CWS males; 54 CWNS females, 62 CWNS males), 8 CWS and 9 CWNS were excluded from this study because they failed to meet the articulation-language criteria. Of the remaining 178 participants, 37 CWS and 49 CWNS were excluded due to incomplete family history of stuttering data across all three time points. An additional 17 more participants were removed because their caregivers did not consistently report a family history of stuttering across all three time points. Thus, the final data corpus consisted of 75 children (25 CWS & 50 CWNS) whose data were analyzed for all but three of the present research questions.

Specifically, for research questions 2 to 4, we additionally excluded children whose caregivers provided inconsistent reports of family histories of articulation disorders ($N = 10$; 4 CWS & 6 CWNS), language disorders ($N = 4$; 3 CWS & 1 CWNS) or ADHD ($N = 8$; 2 CWS & 6 CWNS) across the 3 time points.

Procedures

Each participant and his/her caregiver(s) visited the Vanderbilt Developmental Stuttering Laboratory three times about 8 months apart spanning a period of approximately 16 months. At each time point, during the caregiver interview, the caregiver was presented with a family tree diagram (Richels & Conture, 2010, p.35) and asked to indicate whether the participants' siblings (if applicable), parents, grandparents, and/or great-grandparents stuttered or had other articulation, language, or attention-related disorders as children or adults. A relative was positively identified with a history of one of these issues regardless of whether he/she recovered from the disorder.

For the purposes of the present study, children who were adopted were not included in the final data analysis. Furthermore, to make our findings comparable with previous work in this area (see Table 1), it is important to note that we only analyzed caregiver-report data for the child's sibling(s) and parent(s) (even though caregivers were asked questions about the child's grandparents and great-grandparents). To help the caregiver complete the family tree of disorders, the following verbal instruction was provided by an examiner:

"Here is a diagram that shows a family tree with various emotional, behavioral and communication disorders, with each disorder indicated by letters at the bottom of the page. If you know anyone in your family who has or had any of these disorders, please put – at the

place in the tree (for example, the child's mother) – the letter(s) for the disorder (for example, put AD for attention deficit/hyperactivity disorder). If your child has siblings, you can indicate those on the tree to the right or to the left of the child, using a circle for a girl and a box for a boy.”

Such verbal instructions were repeated for any caregiver who requested them and/or had question(s) about how to complete the family tree of disorders.

Subsequently, the caregiver was asked to report their occupation and highest degree completed and to rate their child's temperamental characteristics on the Behavioral Style Questionnaire (BSQ, McDevitt, & Carey, 1978). While one examiner conducted the parent interview, another engaged with the child during free-play, obtaining at least a 300-word conversational sample (Conture, 2001). Children's stuttering frequency and severity were determined based on these conversational samples, which were analyzed in real-time. After the free-play, children were administered a series of standardized articulation/language tests in the following fixed order: GFTA, PPVT, EVT, and TELD (for additional information about these measures, see above Criteria for articulation, language, and hearing abilities).

Data Analyses

Different statistical procedures were conducted to answer each research question, details of which are presented in the Results below. Of note, according to Bender and Lange (2001), exploratory studies do not require multiple test adjustments. Given the exploratory nature of present study, no adjustment for multiple tests was made.

The main dependent variables for research questions 5 and 6 were measured as follows. Participants' articulation abilities were indexed by GFTA standard scores. Their language abilities were indexed by composite language scores. The composite language scores were calculated by averaging the standard scores from the four standardized language tests (i.e., PPVT, EVT, TELD-receptive, TELD-expressive tests). Their attentional abilities were indexed by the Distractibility scores on the BSQ.

Results

Descriptive Analyses

Consistency of caregiver reports—As mentioned in the method section, of the 92 caregivers who reported a family history of stuttering across 3 consecutive time points, 81.5% ($n = 75$) of them consistently reported either the presence or absence of family history of stuttering in their immediate family members across 3 time points.

Group characteristics: Gender, age, SES & Speech fluency—Prior to testing the main research questions, a series of Fisher's Exact tests assessed the relations between gender and talker groups as well as gender and a family history of stuttering. Additionally, a series of Mann-Whitney U tests (Mann & Whitney, 1947) assessed whether there are differences in chronological age, SES, and speech fluency between CWS and CWNS, as well as between children with a positive versus negative family history of stuttering. Table 2

provides descriptive information of the gender ratio, age, SES, and speech fluency of children (CWS & CWNS) with a positive versus negative family history of stuttering.

Consistent with Yairi and Ambrose (2013), Fisher's Exact Test showed a significant relation between talker groups and gender ($p = .001$, Cramer's $V = .387$), with the CWS sample consisting of more boys ($N = 22$) than girls ($N = 3$), whereas for the CWNS sample, there are relatively equal numbers of boys ($N = 24$) and girls ($N = 26$). There was no significant group difference (CWS vs. CWNS) in chronological age (CWS $M = 44.96$ months, $SD = 6.78$; CWNS $M = 48.78$ months, $SD = 9.61$; $Z = -1.457$, $U = 495.00$, $p = .145$). The SES scores were calculated by multiplying the scale value for occupation (range from 1 to 9) by a weight of 5 and the scale value for education (range from 3 to 7) by a weight of 3 (for more details, see Hollingshead, 2011). There was no significant group difference in SES (CWS $M = 51.22$, $SD = 10.85$; CWNS $M = 50.30$, $SD = 12.11$; $Z = -.236$, $U = 604.00$, $p = .813$). As would be expected based on talker group classification criteria, CWS ($M = 11.91\%$, $SD = 4.57$), compared with their CWNS peers ($M = 4.91\%$; $SD = 2.82$), had significantly more total disfluencies, $Z = -6.034$, $U = 88.500$, $p < .001$. Likewise, CWS ($M = 7.32\%$, $SD = 3.39$), compared with CWNS ($M = 1.37\%$, $SD = .82$) had significantly more stuttered disfluencies, $Z = -7.046$, $U = .000$, $p < .001$. Likewise, CWS had significantly higher mean scores on the SSI-3 ($M = 17.60$; $SD = 4.82$) than CWNS ($M = 6.80$; $SD = 1.86$), $Z = -7.205$, $U = .000$, $p < .001$. Conversely, there was no significant difference in frequency of nonstuttered disfluencies between CWS ($M = 4.59\%$, $SD = 2.96$), and CWNS ($M = 3.54\%$, $SD = 2.71$), $Z = -1.576$, $U = 485.22$, $p = .115$.

Table 2 shows that for all 75 children, there was no significant relation between gender and a family history of stuttering ($p = .757$, Cramer's $V = .048$), although there was a trend that the male to female ratio was higher (18:1) in CWS with a negative family history of stuttering compared to those with a positive family history (2:1), consistent with the findings of Seider et al. (1982). For all 75 children, there was no significant difference in chronological age ($Z = -1.266$, $U = 290.00$, $p = .206$) and in SES ($Z = -.014$, $U = 377.00$, $p = .988$) between children with a positive versus negative family history. Likewise, there were no significant differences in the frequency of stuttered ($Z = -1.609$, $U = 267.00$, $p = .108$), nonstuttered ($Z = -.239$, $U = 361.50$, $p = .811$), total disfluencies ($Z = -.853$, $U = 319.00$, $p = .394$) and SSI total scores ($Z = -1.319$, $U = 289.00$, $p = .187$) between children with a positive (i.e., CWS + CWNS $N = 12$) versus those with a negative (i.e., CWS + CWNS $N = 63$) family history of stuttering. This is consistent with Kidd, Heimbuch, Records, Oehlert, and Webster (1980)'s finding that a family history of stuttering does not appear to be related to severity of stuttering in adults.

Inferential Analyses

Table 6 provides the standardized articulation, language and attention test scores of children (CWS or CWNS or all) with a positive versus negative family history of stuttering. Specific findings based on inferential analyses are discussed below.

Research question 1: Family history of stuttering (CWS vs. CWNS)—For research question 1, Fisher's Exact Test (Mehta & Patel, 1983) assessed the interdependence

between talker groups (CWS vs. CWNS) and a family history of stuttering. Descriptively, more CWS (24%) had a positive family history of stuttering than CWNS (12%). However, the results of the Fisher's Exact Test showed no significant relation between talker groups and a family history of stuttering, ($p = .198$, Cramer's $V = .154$), indicating that CWS and CWNS did not significantly differ in the occurrence of stuttering in immediate families.

Research question 2: Family history of other disorders (CWS vs. CWNS)—For research question 2, three separate Fisher's Exact Tests assessed interdependence between talker groups (CWS vs. CWNS) and family histories of (1) articulation disorders, (2) language disorders and (3) ADHD. Descriptively, 10.5% of CWS and 15.8% of CWNS had a positive family history of articulation disorders. However, the results of the Fisher's Exact Test showed no significant relation between talker groups and a family history of articulation ($p = 1.00$, Cramer's $V = .059$), indicating that CWS and CWNS did not significantly differ in the occurrence of articulation disorders in immediate families. Similarly, CWS and CWNS did not significantly differ in the occurrence of language disorders in immediate families ($p = 1.00$, Cramer's $V = .114$) although informal description of the present sample indicated that 0% of CWS and 4.3% of CWNS had a positive family history of language disorders. Likewise, CWS and CWNS did not significantly differ in the occurrence of ADHD in immediate families ($p = 1.00$, Cramer's $V = .034$) although informal description of the present sample indicated that 9.5% of CWS and 7.3% of CWNS had a positive family history of ADHD.

Research question 3: Family history of stuttering and family history of other disorders in CWS—For research question 3, two separate Fisher's Exact Tests were performed to determine the interdependence between a family history of stuttering and family histories of (1) articulation disorders and (2) ADHD in CWS. Findings indicated that CWS with a positive versus a negative family history of stuttering did not significantly differ in the occurrences of articulation disorders (CWS, $p = 1.00$, Cramer's $V = .205$, Table 3) and ADHD ($p = 1.00$, Cramer's $V = .183$, Table 5) in immediate families. However, the Fisher's Exact Test was not completed to assess the interdependence between a family history of stuttering and family history of language disorders, because there was no CWS with a positive family history of language disorders after excluding 3 CWS with inconsistent reports of family history of language disorders (Table 4).

Research question 4. : Family history of stuttering and family history of other disorders in CWNS—For research question 4, three separate Fisher's Exact Tests were performed to determine the interdependence between a family history of stuttering and family histories of (1) articulation disorders, (2) language disorders and (3) ADHD in CWNS. Findings indicated that CWNS with a positive versus a negative family history of stuttering did not significantly differ in the occurrences of articulation disorders ($p = 1.00$, Cramer's $V = .035$, Table 3) and language disorders ($p = 1.00$, Cramer's $V = .077$, Table 4) in immediate families. However, CWNS with a positive family history of stuttering were more likely to have ADHD in immediate families than CWNS with a negative family history of stuttering ($p = .018$, Cramer's $V = .542$, Table 5). Specifically, 50% of CWNS with a

positive family history of stuttering had a positive family history of ADHD whereas 2.6% of CWNS with a negative family history of stuttering had a positive family history of ADHD.

Research question 5: CWS's family history of stuttering and their articulation, language and attentional abilities—Prior to testing the research questions 5 and 6, we imputed 7 missing values on the TELD receptive and expressive tests using the Expectation Maximization method (Dempster, Laird, & Robin, 1977). These imputed values were used to create the composite language scores (i.e., an index of language abilities) in the analytical model for research questions 5 and 6.

For research question 5, we conducted a series of preliminary analyses to test the normality of distribution and homogeneity of variance to confirm that the assumptions of Analyses of Covariance (ANCOVA; Keselman et al., 1998) are met. Findings indicated that CWS's composite language scores (skewness = $-.142$, Shapiro-Wilk $p = .625$) and BSQ Distractibility scores (i.e., an index of attentional abilities; skewness = $-.219$, Shapiro-Wilk $p = .555$) were normally distributed whereas the distribution of their GFTA standard scores (i.e., an index of articulation abilities) was slightly negatively skewed (skewness = -1.308 , Shapiro-Wilk $p = .037$). Thus, a reflected square root transformation was used to normalize the GFTA data (Quinn & Keough, 2002). After transformation, the distribution of GFTA standard scores became normal (skewness = $.503$, Shapiro-Wilk $p = .673$). Thus, the transformed GFTA scores were used as a dependent variable for research question 5. Also, Levene's test assessed homogeneity of variance and showed that this assumption was not violated for the composite language scores ($p = .835$), transformed GFTA scores ($p = .596$), and BSQ Distractibility scores ($p = .553$).

To address research question 5, three separate ANCOVAs were conducted (Keselman et al., 1998) for only CWS. In each ANCOVA model, groups with a positive versus negative family history within CWS served as an independent variable and participants' (1) transformed GFTA scores, (2) composite language scores, or (3) BSQ Distractibility scores served as dependent variables respectively. The following covariates were included in the model to control for their possible effects on the dependent variables: participants' chronological age (in months) at the first visit, gender and maternal education (Dollaghan et al., 1999).

For CWS, the results of ANCOVA indicated no main effect of a family history of stuttering for articulation ($F = 2.991$, $p = .099$, $\eta^2 = .130$, observed power = $.377$), language ($F = .965$, $p = .338$, $\eta^2 = .046$, observed power = $.155$), and attentional abilities ($F = .642$, $p = .433$, $\eta^2 = .034$, observed power = $.118$) (see Table 6).

Research question 6: CWNS's family history of stuttering and their articulation, language and attentional abilities—For research question 6, we conducted a series of preliminary analyses to test the normality of distribution and homogeneity of variance to confirm the assumptions of ANCOVA are met. Findings indicated that the BSQ Distractibility (skewness = $.264$, Shapiro-Wilk $p = .124$) and GFTA standard scores (skewness = $-.451$, Shapiro-Wilk $p = .421$) were normally distributed whereas the composite language scores were significantly negatively skewed (skewness = $-$.

804, Shapiro-Wilk $p = .031$). Thus, a reflected square root transformation was used to normalize the composite language scores (Quinn & Keough, 2002). After transformation, the distribution of composite language scores became normal (skewness = $-.315$, Shapiro-Wilk $p = .668$). Thus, the transformed language composite scores were used as a dependent variable for research question 6. Also, Levene's test assessed homogeneity of variance and showed that this assumption was not violated for the transformed composite language scores ($p = .733$), GFTA standard scores ($p = .336$), and BSQ Distractibility scores ($p = .537$).

To address research question 6, three separate ANCOVAs were conducted for CWNS only. In each ANCOVA, groups with a positive versus negative family history within CWNS served as an independent variable and participants' (1) GFTA standard scores, (2) transformed composite language scores, or (3) BSQ Distractibility scores served as dependent variables respectively. Participants' chronological age (in months) at the first visit, gender and maternal education were covariates.

For CWNS, the results of ANCOVA indicated no main effect of a family history of stuttering for articulation ($F = .566$, $p = .456$, $\eta^2 = .012$, observed power = $.114$) and attentional abilities ($F = 1.339$, $p = .254$, $\eta^2 = .032$, observed power = $.204$). However, results of ANCOVA indicated a significant main effect of a family history of stuttering on children's language abilities ($F = 10.848$, $p = .002$, $\eta^2 = .194$, observed power = $.897$) (see Table 6). Specifically, CWNS with a positive family history of stuttering exhibited lower language abilities than those with a negative family history of stuttering.

Discussion

Overview of Main Findings

Findings indicated that (1) a relatively high proportion (81.5%) of caregivers reported consistently a positive or negative family history of stuttering across time, (2) CWNS who had a positive family history of stuttering were more likely to report a family history of ADHD than CWNS with a negative family history of stuttering, and (3) CWNS who had a positive family history of stuttering had lower language abilities than those with a negative family history of stuttering. The general implications of each of these findings as well as other non-significant findings will be discussed below.

Consistent Report of Family History of Stuttering Over Time

The first finding indicated that a relatively high proportion (i.e., 81.5%) of caregivers *consistently* reported a positive or negative family history of stuttering across three consecutive time points. Although caregiver reports have been routinely used to determine a child's family history of stuttering, the reliability of such methods have not always been clearly reported. In attempts to address this concern, we employed a relatively strict verification procedure. Our finding should provide some degree of assurance to those who collect such caregiver reports, whether for clinical or research purposes. We speculate that the inconsistent reports of the remaining 18.5% of caregivers may have resulted from later changes or correction of information. For example, by the second or third time point, additional family members may have developed or recovered from stuttering, or caregivers

may have later learned that their initial information was inaccurate. Perhaps, some inconsistent reports related to informants' reluctance to share their family's health profile with unfamiliar examiner. Nevertheless, the fact that the majority of parents appear consistent in reporting a family history of stuttering suggests that those who collect such information can assume that such reports are reasonably stable over time.

Relation of Attentional Vulnerability to Family History of Stuttering

The second main finding indicated that CWNS with a positive family history of stuttering were more likely to have a positive family history of ADHD than CWNS with a negative family history of stuttering. Such findings are curious given that (1) no significant relation was found between CWS's family history of stuttering and family history of ADHD (a finding associated with research question 3); and (2) children with a positive versus negative family history of stuttering did not significantly differ in their attentional abilities based on caregiver reports (findings associated with research questions 5 and 6). Nevertheless, the present finding indicates a possible association between ADHD and stuttering at a genetic or epigenetic¹ level. This speculation is supported by comorbidity findings of ADHD and stuttering, ranging from 4% (Arndt & Healey, 2001) to 26% (Riley & Riley, 2000). Donaher and Richels (2012) further reported that both disorders have a higher concordance in identical than fraternal twins (Andrews, Morris-Yates, Howie, & Martin, 1991; Godai, Tatarelli, & Bonanni, 1976) and occur more often in boys than girls. Regarding the association among stuttering, attention, and gender, Clark et al. (2015) found that "hyper-attentiveness" or minimal distractibility (i.e., minimal rapid shifting and/or fluctuating of attentiveness) is more frequent in young boys than young girls who stutter, consistent with the results of an earlier study by Anderson, Pellowski, and Conture (2003). Interestingly, neuroimaging studies have shown that both individuals with ADHD (Aylward et al., 1996; Hart, Radua, Nakao, Mataix-Cols, & Rubia, 2013; Teicher et al., 2000) and those who stutter (Alm, 2004; Giraud et al., 2008; Tani & Sakai, 2011) have structural and functional differences in the *basal ganglia* compared to the typical population. In the same vein, findings from genetic studies suggested that dopamine genes are associated with both ADHD (Swanson et al., 2000) and stuttering (Lan et al., 2009). Further investigations are warranted to better understand the possible association between stuttering and ADHD, employing different or advanced methodologies than those used in the present study.

Relation of Linguistic Vulnerability to Family History of Stuttering

The third main finding indicated that CWNS with a positive family history of stuttering had lower language abilities than those with a negative family history of stuttering. Although young CWNS exhibited a significant association between children's language abilities and caregiver reports of a family history of stuttering, CWS exhibited no such association. This non-significant finding for CWS is not completely surprising because it is consistent with Seider et al.'s (1982) finding based on adults who stutter. On the other hand, the present null finding for CWS may have been attributed to lower observed power for CWS when compared to CWNS. Thus, further research with a larger sample size is warranted to more

¹Epigenetic refers to "a heritable state of gene expression that is not due to changes in the DNA sequence." (Barres & Zierath, 2011, p.899)

adequately determine the association between CWS's language abilities and family history of stuttering.

That being said, findings suggest that a possible linguistic vulnerability may be transmitted in families with a positive history of stuttering. Such a linguistic phenotype² of children with a positive family history of stuttering may be a manifestation of several underlying factors: (1) genetics or epigenetics, (2) environment, and/or (3) gene-environment interaction (Riches et al., 2011). With the current data, however, it is challenging to determine whether/how these underlying factors contribute to linguistic vulnerabilities among CWNS with a positive family history of stuttering. Thus, further investigation of genetic, epigenetic or environmental contributions to linguistic traits is warranted to determine how children's linguistic vulnerability may be associated with familial and/or genetic predisposition to stuttering.

With the above cautions in mind, the following speculation is put forward. Perhaps, genetic predispositions to stuttering are associated with several vulnerabilities in emotional, linguistic, motoric, or other processes. Depending on the weighting of each vulnerability, and interactions with other (e.g., environmental) stimuli/stressors, a child's genetic predisposition to stuttering may or may not result in the onset of stuttering. Such a speculation is based on the multifactorial model of stuttering (Smith & Kelly, 1997), suggesting that no single factor is necessary or sufficient for stuttering to occur. Instead, the model suggests that it is "the weighting of factors that determines whether an individual is in the diagnostic space of stuttering" (p.209). Similarly, Yairi and Seery (2015) also suggested that "a particular characteristic that increases the susceptibility for stuttering may not, by itself, cause the stuttering, but when it *co-occurs* with certain other characteristics, stuttering may be expressed" (p.167). Thus, linguistic vulnerability may be a component of a group of vulnerabilities in a family with a history of stuttering.

Talker Group and Family Histories of Stuttering, Articulation Disorders, Language Disorders and ADHD

Research questions 1 and 2 of the present study produced null findings that warrant discussion. Regarding the research question 1, although descriptive statistics suggested more caregiver reports of a family history of stuttering among CWS than CWNS, inferential statistics indicated no significant relation between a family history of stuttering and talker groups. Such unexpected findings are not surprising for the following reasons. First, many previous empirical studies on young children's stuttering reported descriptive but inferential analyses, making it difficult to compare present findings. Second, as shown in Table 1, our descriptive findings fall within the range reported in the literature. Specifically, our study found that (1) 24% of CWS had a family history of stuttering, compared to 23.3% to 72.1% reported in the literature; and (2) 12% of CWNS had a family history of stuttering, compared to 6% up to 15.6% reported in the literature. Discrepant findings between the present and previous studies may be due to differences in methodologies (e.g., obtaining data from multiple vs. single informants) and participant characteristics (e.g., extended vs.

²A phenotypic trait is an "observable manifestation of an underlying genetic code, or a gene-environment interaction" (Riches, Loucas, Baird, Charman, & Simonoff, 2011, p.24).

immediate families, persistent vs. recovered stutterers; for further details pertaining to these between-study methodologies, see Table 1). For example, we found comparable results upon comparing the present study to one particular study with participants of similar characteristics (e.g., ages 2 to 6 years; Shin, Chon, and Lee, 2011, with 27.3% of young Korean children (N = 227) having first-degree relatives who stuttered compared to the 24% of children in the present study. Fourth, perhaps our relatively lower percentage of CWS with a positive family history of stuttering resulted from the present study's rigorous selection criteria (i.e., excluding participants if caregiver reports were inconsistent across three time points over the course of 16 months). For example, Ambrose, Yairi and Cox (1993), updated/added new cases of stuttering in families across four time points. Some of these "updated" or "additional" families would have been excluded from the present study because they did not meet the "consistency criteria". As would be recalled, we excluded 17 children (9 CWS and 8 CWNS) from the final data corpus due to the consistency criteria. When we added those 17 children back to the data sample, the percentage of CWS with a positive family history rose to 42.4%. These updated CWS findings are comparable to those of Ambrose et al. who reported that 43% CWS had a positive family history of stuttering in their immediate family. We await future investigations to determine the reliability and validity of these varying subject selection procedures.

The finding related to the research question 2 indicated no significant relations between talker groups and family histories of 1) articulation disorders, 2) language disorders, and 3) ADHD. Although Neils and Aram (1986) reported that children with *language disorders* had significantly more family histories of *stuttering* as well as speech, reading and language disorders than the control group, the present finding does not support such cross-disorder familial risk. Specifically, this finding suggests that familial vulnerability to articulation, language or attention related to disorders *per se* does not appear to contribute to onset and development of stuttering. Further investigation on cross-disorder familial risk is warranted to determine how children's genetic, epigenetic, or environmental vulnerability to articulation, language and attention related disorders is associated with onset and development of stuttering.

Limitations

Several limitations are worth mentioning when interpreting the present study's findings. First, the present methods did not involve genetic analyses that would have allowed us to more directly examine genetic contributions to children's speech-language. Still, this exploratory investigation did use careful, replicable inclusion criteria, with findings contributing to a comprehensive understanding of childhood stuttering.

Second, although the number of generations per participant was a priori controlled, other factors such as the number of siblings per participant were not. Hence, the present data do not allow us to determine the role played, if any, by the number of siblings in either reported a family history of stuttering or other participant characteristics (e.g., level of language development). Future studies should consider obtaining information regarding the number and gender of participants' siblings.

Third, both CWS's and CWNS's sample sizes were relatively modest suggesting low to high power to reject the null hypothesis (see the observed power reported in the results related to research questions 5 and 6).

Fourth, although rare, it is possible that some caregivers of CWNS with a positive family history of stuttering participated in the present study because they wanted to make sure that their children are not at risk of developing stuttering due to their positive family history of stuttering. If this were the case, this might have contributed to an inflation of CWNS with a positive family history of stuttering, which may account for the present finding of no significant difference in a family history of stuttering between CWS and CWNS. Similarly, it is also possible that some caregivers of CWNS with a positive family history of stuttering might notice their children's speech-language and/or behavioral problems more readily than those without a family history of stuttering, which might have led them to bring their children to the laboratory for this study.

Fifth, family history data were obtained mostly from a single informant rather than multiple informants. This may have increased false-positive or false-negative identification of family histories of disorders especially regarding the informant's spouse's side of family. Nonetheless, this may make our findings more applicable to clinical practice and/or research studies because in most clinical settings, speech-language pathologists obtain family history data from a single informant (for practical reasons, such as the availabilities of both caregivers). That being said, future studies might consider collecting family history information from at least two informants as suggested by Ambrose et al. (1997) to increase reliability of the data.

Conclusion

Overall, the present findings show that caregiver reports of a family history of stuttering are reasonably stable over time. Furthermore, findings provide an insight into the nature of vulnerabilities that is transmitted in families with a positive history of stuttering. Specifically, findings may be cautiously taken to suggest a possible association between a family history of ADHD and a family history of stuttering at least for CWNS. Questions about whether the nature of such associations is genetic, environment or their interaction must await further study for answers. Similarly, relatively lower language abilities during early childhood may be observed in families with a history of stuttering. Perhaps, such vulnerability in language may contribute to a child's stuttering depending on whether it co-occurs with other vulnerabilities or environmental stressors.

Taken together, future investigations are warranted to determine possible linguistic and attentional vulnerabilities in children with a positive family history of stuttering. Such studies should consider using more cortical, experimental and genetic methodologies to further elucidate and elaborate on present findings.

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Highlights

1. Caregiver reports of a family history of stuttering are reasonably stable over time.
2. CWNS with a family history of stuttering tended to have a family history of ADHD.
3. CWNS with a family history of stuttering exhibited relatively low language ability.
4. Results shed light on the vulnerabilities in families with a history of stuttering.

Table 1

Twenty one selected accessible studies (from 1937 to 2011) of family histories of stuttering – alphabetically listed – for people who do (PWS) and do not stutter (PWNS) across varying ages. Percentages of immediate and extended family members who stutter are provided whenever available.

Study	Participants			PWS With a Family History of Stuttering (%)	PWNS With a Family History of Stuttering (%)	Informants
	PWS (n)	PWNS (n)	Age (years)			
Ambrose, Yairi, & Cox (1993)	69	-	2–6	43% in immediate; 71% in extended	-	Both parents when possible
Andrews & Harris (1964)	80	80	9–11	37.5%	1.3%	Mothers
Bryngelson (1939)	78	78	17–31	54%	6%	Self
Bryngelson & Rutherford (1937)	74	74	4–16	46%	18%	Parents & relatives
Buck, Lees, & Cook (2002)	61	-	2–6	45.9% in immediate; 72.1% in extended	-	Both parents when possible
Cooper (1972)	187	-	12–17	28%	-	Self
Darley (1955)	50	50	2–14	52%	42%	Both parents
Drayna, Kilshaw, & Kelly (1999)	1999 (persistent)	-	>14	53.3% of persistent	-	Self
Johnson & Associates (1959)	150	150	2–8	23.3%	6%	Parents
Mansson (2000)	12 (persistent) ¹	-	5	50% in immediate; 67% in extended	-	Parents
Martyn & Sheehan (1968)	37 (persistent); 48 (recovered)	277	16–56	32.9% of total N (40.5% of persistent; 27.1% of recovered);	6.1%	Self
Meyer (1945)	100	246	10–50	61%	6.5%	-
Porfert & Rosenfield (1978)	44 (persistent); 60 (recovered)	1965	college students	21.2% of total N (29.5% of persistent; 15% of recovered);	5%	Self
Poulos & Webster (1991)	169	-	14–60	46% in immediate; 66% in extended	-	Self
Shin, Chon, & Lee (2011)	277	-	2–6	27.3% in immediate; 49.3% in extended	-	Parents
Viswanath, Lee, & Chakraborty (2004)	56	-	adults	84% in extended	-	Self, parents or spouses
Wepman (1939)	250	250	children	68.8%	15.6%	Parents
West, Nelson, & Berry (1939)	204	204	4–30	51% in extended	18.1% in extended	N/A
Yairi (1983)	22	-	2–3	45% in immediate; 64% in extended	-	Mothers
Yairi & Ambrose (1992)	87	-	1–6	46.6% in immediate; 66.3% in extended	-	Parents
Yairi & Ambrose (2005)	123	-	children	69% of total N (88% of persistent; 65% of recovered)	-	Parents

Study	Participants			PWS With a Family History of Stuttering (%)	PWNS With a Family History of Stuttering (%)	Informants
	PWS (n)	PWNS (n)	Age (years)			
MEAN				Immediate: 43.4% Extended: 65.6% Persistent: 52.8% Recovered: 35.7% Unspecified: 46.3%	Extended: 18.1% Unspecified: 11.8%	

Although 1,021 children participated in the study, family history data was only reported for a subset of children whose stuttering persisted for two years and provided family history data (n=12). Thus, data reported in this table (e.g., age of participants and percentages of family members who stuttered) are for that subset.

Note: N/A = Not available due to limited access to the original articles.
Immediate = participant's family including parents and siblings (and offspring if participants were adults);
Extended = participant's family including parents and siblings, grandparents, aunts, uncles, and other relatives.
Persistent = individuals with persistent stuttering; Recovered = individuals who recovered from stuttering

Descriptive information for the 75 participants – children who stutter (CWS; n = 25) and children who do not stutter (CWNS; n = 50) – whose data were included in this study's final data corpus.

Table 2

Measures	Children with a <i>positive</i> family history of stuttering			Children with a <i>negative</i> family history of stuttering		
	All children (CWS+ CWNS, n=12)	CWS (n=6)	CWNS (n=6)	All children (CWS+ CWNS, n=63)	CWS (n=19)	CWNS (n=44)
Gender (Male:Female)	2:1	2:1	2:1	1.52:1	18:1	1:1.2
Age (in months)	44.46 (7.68)	45.13 (7.79)	43.78 (8.23)	48.09 (9.06)	44.91 (6.67)	49.46 (9.67)
SES (range from 14 to 66)	52.67 (4.89)	54.33 (6.23)	51.00 (2.66)	50.21 (12.51)	50.24 (11.91)	50.20 (12.89)
Stuttered Disfluencies (SD, %)	4.08 (2.93)	6.39 (2.28)	1.78 (.96)	3.21 (3.59)	7.61 (3.68)	1.31 (.80)
Nonstuttered Disfluencies (ND, %)	3.39 (1.72)	3.00 (2.19)	3.78 (1.15)	3.98 (2.99)	5.09 (3.05)	3.51 (2.87)
Total Disfluencies (TD, %)	7.47 (2.85)	9.39 (2.55)	5.56 (1.60)	7.20 (5.11)	12.70 (4.82)	4.82 (2.95)
Stuttering Severity (SSI-3)	11.67 (6.01)	16.00 (3.35)	7.33 (2.07)	10.59 (6.15)	18.11 (5.17)	6.73 (1.85)

* Note.

SD=stuttered disfluencies

NSD=non-stuttered disfluencies

TD=total disfluencies SSI-3=Stuttering Severity Instrument-3 (Riley, 1994)

SES (social-economic status) was determined by Hollingshead's four factor index (2011), which ranged from 14 to 66, with lower values indicating lower SES.

Family history of stuttering = Each participant's a positive (i.e., presence) or negative (i.e., absence) family history of stuttering was based on his or her caregiver's consistent report across 3 consecutive time points (8 months apart) spanning a total of 16 months.

Table 3

For CWNS (n=44) and CWS (n=21) with consistent reports of a family history of articulation disorders (AD), the relations between a family history of stuttering and a family history of articulation disorders (AD).

	CWNS with a family history of stuttering (n=6)	CWNS (n=44) CWNS without a family history of stuttering (n=38)	Fisher's Exact Test p value
CWNS with a family history of AD	1	5	p=1.00
CWNS without a family history of AD	5	33	
	CWS with a family history of stuttering (n=6)	CWS (n=21) CWS without a family history of stuttering (n=15)	Fisher's Exact Test p value
CWS with a family history of AD	0	2	p=1.00
CWS without a family history of AD	6	13	

Table 4

For CWNS (n=49) and CWS (n=22) with consistent reports of a family history of language disorders (LD), the relations between a family history of stuttering and a family history of LD.

	CWNS with a family history of stuttering (n=6)	CWNS (n=49) CWNS without a family history of stuttering (n=43)	Fisher's Exact Test p value
CWNS with a family history of LD	0	2	p=1.00
CWNS without a family history of LD	6	41	
	CWS with a family history of stuttering (n=6)	CWS (n=22) CWS without a family history of stuttering (n=16)	Fisher's Exact Test p value
CWS with a family history of LD	0	0	N/A
CWS without a family history of LD	6	16	

Note. N/A = non-applicable. No statistics were computed because there were not two variables.

Table 5

For CWNS (n=44) and CWS (n=23) with consistent reports of family history of ADHD, the relations between a family history of stuttering and a family history of ADHD.

	CWNS with a family history of stuttering (n=4)	CWNS (n=44) CWNS without a family history of stuttering (n=40)	Fisher's Exact Test p value
CWNS with a family history of ADHD	2	1	p=.018
CWNS without a family history of ADHD	2	39	
	CWS with a family history of stuttering (n=6)	CWS (n=22) CWS without a family history of stuttering (n=16)	Fisher's Exact Test p value
CWS with a family history of ADHD	0	2	p=1.00
CWS without a family history of ADHD	6	15	

Table 6

Means and Standard Deviations (SD) for measures of attention, articulation and language abilities for all participants (N = 75), young CWS (n=25) and CWNS (n=50) with a positive versus negative family history of stuttering.

Measures	Children with a positive family history of stuttering			Children with a negative family history of stuttering		
	All children (CWS+, CWNS, n=12)	CWS (n=6)	CWNS (n=6)	All children (CWS+, CWNS, n=63)	CWS (n=19)	CWNS (n=44)
Attentional abilities						
	3.70 (.57)	3.76 (.78)	3.64 (.34)	3.90 (.06)	3.66 (.57)	4.01 (.59)
Articulation abilities						
	112.75 (8.34)	115.50 (4.89)	110.00 (10.53)	111.10 (8.40)	109.42 (8.78)	111.82 (8.23)
	113.08 (13.72)	120.50 (11.42)	105.67 (12.32)	118.52 (10.91)	115.37 (10.42)	119.89 (10.96)
Language abilities						
	117.17 (9.33)	120.33 (12.37)	114.00 (3.80)	118.89 (10.60)	115.16 (8.85)	120.50 (10.97)
	118.25 (17.05)	120.67 (19.41)	115.83 (15.77)	121.68 (13.78)	123.39 (12.78)	120.95 (14.27)
	108.67 (16.03)	114.17 (18.25)	103.17 (12.64)	115.08 (13.65)	116.17 (19.36)	114.61 (10.50)
Language composite scores	114.29 (11.43)	118.92 (12.38)	109.67 (9.08)	118.42 (8.45)	117.25 (8.71)	118.92 (8.38)

* Note.

GFTA="Sounds in Words" subtest of the *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); PPVT=Peabody Picture Vocabulary Test-4 (Dunn & Dunn, 2007); EVT=Expressive Vocabulary Test-2 (Williams, 2007); TELD=Test of Early Language Development-3 (Hresko, Reid, & Hamill, 1999); TELD receptive scores were based on 24 CWS and 48 CWNS, and TELD expressive scores were based on 24 CWS and 47 CWNS due to incomplete data; BSQ= Behavioral Style Questionnaire (McDevitt, & Carey, 1978), the higher the parent report of the child's Distractibility, the less the child is considered to be distracted; Participants' Distractibility score was based on 23 CWS and 46 CWNS due to incomplete data.