LSHSS

Research Article

Developmental Stuttering in Children Who Are Hard of Hearing

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Purpose: A number of studies with large sample sizes have reported lower prevalence of stuttering in children with significant hearing loss compared to children without hearing loss. This study used a parent questionnaire to investigate the characteristics of stuttering (e.g., incidence, prevalence, and age of onset) in children who are hard of hearing (CHH).
Method: Three hundred three parents of CHH who participated in the Outcomes of Children With Hearing Loss study (Moeller & Tomblin, 2015) were sent questionnaires asking about their child's history of stuttering.
Results: One hundred ninety-four parents of CHH responded to the survey. Thirty-three CHH were reported to have stuttered at one point in time (an incidence of 17.01%), and 10 children were still stuttering at the time of survey submission (a prevalence of 5.15%). Compared to estimates in the general population, this sample displayed a significantly higher incidence and prevalence. The age of onset, recovery rate, and other characteristics were similar to hearing children.
Conclusions: Based on this sample, mild to moderately severe hearing loss does not appear to be a protective factor for stuttering in the preschool years. In fact, the incidence and prevalence of stuttering may be higher in this population compared to the general population. Despite the significant speech and language needs that children with mild to moderately severe hearing loss may have, speech-language pathologists should appropriately prioritize stuttering treatment as they would in the hearing population.
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Stuttering is a communication disorder characterized by involuntary disruptions in the smooth forward flow of speech. Fluency lies along a continuum in which one end represents completely smooth forward flowing speech and the other end characterizes speech that is not moving forward at all. It is not uncommon for all speakers, especially young children whose speech and language skills are still developing, to occasionally exhibit "normal" disfluencies like interjections, phrase repetitions, or multisyllabic whole-word repetitions. However, most speakers rarely have "stuttering-like disfluencies" in the form of sound or syllable repetitions, audible or inaudible sound prolongations, or monosyllabic whole-word repetitions (Ambrose & Yairi, 1999). When a person exhibits a high percentage of stuttering-like disfluencies, typically 3% of syllables, they are classified as a person who stutters. This criterion has recently been questioned for multilingual speakers, as Byrd, Bedore, and Ramos (2015) found that bilingual speakers who do not stutter exhibit higher levels of stuttering-like disfluencies (with the exception of prolongations) than monolingual children who do not stutter. *Developmental stuttering* is a term used to describe stuttering behavior that first emerges in the early childhood years, typically following a time when the child was previously speaking fluently (meaning very few or no stuttering-like disfluencies; Bloodstein & Ratner, 2008). In this article, we describe a preliminary investigation of the incidence and prevalence of developmental stuttering in young children with mild to moderately severe hearing loss, from this point on referred to as children who are hard of hearing (CHH).

Developmental Stuttering in Typically Hearing Children

The combined fruits of several epidemiological studies (Buck, Lees, & Cook, 2002; Craig, Hancock, Tran, Craig, & Peters, 2002; Howell, Davis, & Williams, 2008; Månsson, 2000, 2005; Proctor, Yairi, Duff, & Zhang, 2008; Reilly et al., 2009) and longitudinal studies (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Smith, Goffman, Sasisekaran, & Weber-Fox, 2012; Weber-Fox, Wray, & Arnold, 2013;

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Yairi & Ambrose, 1992, 1999) over the last quarter century have provided improved understanding of the characteristics of developmental stuttering, including incidence and prevalence, age of onset, and the rate of recovery or persistence. Yairi and Ambrose (2013) synthesized the current knowledge of the incidence and prevalence of stuttering in a review where they noted that developmental stuttering typically begins during the preschool years between the ages of 2 and 4 years, with over 75% of children "outgrowing" stuttering within 18 months of onset. The incidence of stuttering (i.e., the percentage of people who have ever exhibited stuttering behavior in their lifetime) is between 5% and 8% of the general population. Prevalence of stuttering (i.e., the percentage of people who are currently exhibiting stuttering behavior) is estimated to be approximately 1%. Of the 1% who are currently stuttering, the vast majority are preschool aged, and most will not persist in stuttering into adulthood (Yairi & Ambrose, 2013).

It is hypothesized that the onset and development of stuttering is multifactorial, meaning that there are many factors that likely contribute to this, including genetics, speech, language, temperamental, and environmental factors (Conture & Walden, 2012; Smith & Weber, 2016). In spite of the complex and dynamic interaction of factors that contribute to stuttering, recent studies investigating preschoolers who stutter close to the age of onset, especially longitudinal studies, have identified potential speech and language factors that may contribute to stuttering onset and persistence. Although there is a great deal of heterogeneity in speech and language abilities of children who stutter, general trends are also observed. For example, a meta-analysis of overall language abilities found that children who stutter scored significantly lower on standardized tests of overall language measures (including separate receptive and expressive scores), as well as mean length of utterance, compared to typically fluent children (Ntourou, Conture, & Lipsey, 2011). There is substantial evidence that preschoolers who stutter have a higher prevalence of phonological or articulation disorders (Arndt & Healey, 2001; Blood, Ridenour, & Qualls, 2003; Blood & Seider, 1981; Wolk, Edwards, & Conture, 1993). Longitudinal studies have shown that, close to the onset of stuttering, children who persist show poorer phonological and articulation abilities (Paden, Ambrose, & Yairi, 2002; Spencer & Weber-Fox, 2014) as well as nonword repetition performance (Spencer & Weber-Fox, 2014) compared to children who recover from stuttering. However, no single variable provides predictive ability to differentiate at the individual level that children will or will not recover from stuttering. At present, the data suggest that these factors should be viewed as potential risk factors and should be weighed together with other potential factors like language dissociations (Anderson, Pellowski, & Conture, 2005; Clark, Conture, Walden, & Lambert, 2015; Coulter, Anderson, & Conture, 2009), family history of stuttering (Kraft & Yairi, 2012), and the goodness of fit between temperament and environmental contexts (Ambrose et al., 2015; Jones, Choi, Conture, & Walden, 2014).

Relationship Between Stuttering and Audition

Many phenomena that tend to reduce the frequency of stuttering involve changes in audition. For example, delayed auditory feedback, frequency shifted feedback, shadowing another person's speech, and noise that blocks the speaker from hearing his or her own voice temporarily increase fluency in some, but not all, persons who stutter (Bloodstein & Ratner, 2008). The mechanisms that underlie improved fluency with altered audition are not fully understood, but some have hypothesized that people who stutter rely too heavily on sensory feedback during speech production (Hutchinson & Ringel, 1975; Tourville, Reilly, & Guenther, 2008; van Lieshout, Peters, Starkweather, & Hulstijn, 1993). Civier, Tasko, and Guenther (2010) used the Directions Into Velocities of Articulators neural network model to demonstrate how an increased reliance on auditory feedback could result in disfluent behavior, specifically repetitions. Most pertinent to this study is that the feedback-biased model produced less disfluencies during simulations with auditory masking, with a greater reduction in stuttering when the masking was louder. It was hypothesized that masking increases fluency because it increases the signal-to-noise ratio, making it less likely that errors will be detected between the intended and actual speech signal. They further hypothesized that "Hearing loss and whispering are additional conditions that affect the intensity (as well as quality) of auditory feedback, and according to the current account, should improve fluency as well. Indeed, the incidence of stuttering, in children who are deaf or hard of hearing, is low, and especially so in the completely deaf" (p. 265). This model highlights why it may be theoretically important to investigate the relationship between stuttering and the degree of hearing loss. It may be that significant hearing loss does indeed reduce the prevalence of stuttering (Backus, 1938; Harms & Malone, 1939; Montgomery & Fitch, 1988), but more data are needed regarding stuttering in CHH to determine the relationship between audition and stuttering across the spectrum of hearing loss. Investigating CHH may provide further insight into the association between audition and stuttering and further refinement of current theoretical models.

Developmental Stuttering in Children With Hearing Loss

Across individuals from diverse backgrounds (e.g., race and cultures), similar incidence and prevalence rates of stuttering have been found (Bloodstein & Ratner, 2008). One exception is the low prevalence of stuttering that has been reported in children with hearing loss. Three studies using teacher surveys sent to schools for the deaf/hard of hearing found extremely low occurrences of stuttering from significant sample sizes (Backus, 1938; Harms & Malone, 1939; Montgomery & Fitch, 1988). Two early studies had a combined sample of 28,000 students from "schools for the deaf," and there was a report of only 14 children who stuttered, a prevalence of about 0.05% (Backus, 1938; Harms

& Malone, 1939). A more recent study used similar survey methodology from a sample of 9,930 children from schools for the deaf/hard of hearing. Only 12 children exhibited stuttering behavior, a prevalence of about 0.12% (Montgomery & Fitch, 1988). It is important to note that in this study three children stuttered in the oral mode, six children stuttered in the manual mode, and three children stuttered in both modes. Given that the prevalence of stuttering in school-age children is estimated to be at least 1% (Yairi & Ambrose, 2013), the low prevalence reports in children with hearing loss are quite striking.

Despite large sample sizes, the three studies that investigated stuttering in the deaf/hard of hearing population are limited in several ways. First, the studies used teacher surveys that asked about children's stuttering during the school-age years, a period when most children have recovered from stuttering. The teachers likely would not have had knowledge of the children's stuttering during the preschool years when stuttering is more prevalent. A similar survey study would be better aimed at parents who had first-hand experience with the child during the preschool years. Second, two of the studies were not specific in reporting the degree of hearing loss of the sampled children (Backus, 1938; Harms & Malone, 1939). Given that most of the children were enrolled in a self-contained school for the deaf or hard of hearing, we may conclude that the participants had severe to profound hearing loss, and children with mild to severe hearing loss were underrepresented. CHH are typically educated in the regular education setting (Karchmer, Mitchell, Marschark, & Spencer, 2003). Third, the studies did not collect speech or language data to help identify how these factors may have been related to stuttering in this population.

Speech and Language Abilities of Children With Mild to Moderate Hearing Loss

CHH are a historically underserved and underresearched population (Donahue, 2007). The lack of research has resulted in limited knowledge regarding developmental outcomes and susceptibility to a range of communication disorders in CHH (Eisenberg et al., 2007). This gap in the research literature has been rectified in recent years, with the implementation of several longitudinal research studies (Ching et al., 2013; Holte et al., 2012; Stika et al., 2015). The Outcomes of Children With Hearing Loss (OCHL) project is one of these multicenter, longitudinal studies. The goal of the project was to obtain a clearer understanding of the speech, language, and auditory outcomes of CHH who rely on spoken English in the absence of additional disabilities (Moeller & Tomblin, 2015). Recent evidence from the OCHL team indicates that hearing loss in the mild to moderately severe range (i.e., 25–75 dB HL) places these children at risk for speech and language delays compared to their peers with typical hearing (Tomblin, Harrison, Ambrose, Oleson, & Moeller, 2015). These delays are most evident in the domain of morphosyntax, although vocabulary and articulation skills are also depressed during the preschool years. Auditory access, through the consistent use of early, well-fit hearing aids, appears to mitigate the challenges of hearing loss for this population (Moeller, Tomblin, & OCHL Collaboration, 2015). Despite the recent emergence of longitudinal studies of CHH to investigate speech and language outcomes, to our knowledge there has not been a systematic investigation of stuttering in this population to compare to previous studies that investigated stuttering in populations with more severe hearing losses.

Purpose of the Study

The unanswered questions regarding stuttering in CHH are important given the predominant view that stuttering is less prevalent in people with hearing loss (Andrews et al., 1983; Montgomery & Fitch, 1988). A better understanding of how hearing loss relates to stuttering is important both theoretically (e.g., the role of auditory feedback in stuttering) and clinically (e.g., informing clinicians who work with CHH). This study examines characteristics of stuttering in CHH via parent reports during the preschool years. If stuttering behaviors were reported, parents provided information about stuttering onset, at what age the child stopped stuttering (if they had stopped), characteristics of the stuttering, and whether the child had ever been treated for stuttering. Longitudinal data from the OCHL study (Tomblin, Walker, et al., 2015) allowed for investigation of the relationship between stuttering and speech, language, and hearing variables. Our research questions are indicated below.

1. What is the incidence and prevalence of stuttering in CHH?
2. What are the characteristics of stuttering in CHH (e.g., age of onset and types of disfluencies)?
3. In CHH, around the average age of stuttering onset, are there differences in the speech, language, or hearing abilities between children with and without a history of stuttering, and are there differences between children who recovered from stuttering versus those who are at risk of persisting?

Method

Participants

Participants were parents of CHH who were recruited to participate in the OCHL longitudinal study (Tomblin, Walker, et al., 2015). Children with a confirmed sensorineural, mixed, or permanent conductive bilateral hearing loss between 25 and 75 dB HL were included. All had at least one parent or primary caregiver who spoke English in the home. Children with developmental disabilities in addition to hearing loss were not included. Families were recruited from three study sites and surrounding states: University of Iowa, Iowa City; Boys Town National Research Hospital, Omaha, Nebraska; and University of North Carolina, Chapel Hill. For the OCHL study, approval was obtained from the institutional review board at each research center.

For the fluency survey, institutional review board approval was obtained from the University of Iowa.

Requests were sent to 303 parents of children in the OCHL study to respond to a fluency survey to obtain information about their children's history of stuttering. Two hundred and seventy-five of the requests were e-mailed to the parents with a link to an online version of the survey and a unique identifying number that tied their response to their child's OCHL data. E-mail addresses were not available for 28 of the parents, so a paper copy of the survey was sent to their home address with a postage-paid return envelope. There were 194 responses to the survey, 181 online responses and 13 paper copies, for a return rate of 64%. The 13 hard copy surveys that were mailed in were entered into the online survey system by two different research assistants to ensure reliable transfer of data. Survey responses were received in the spring and summer of 2015. Respondents were not compensated for their participation in the survey, but the high return rate is likely related to the brief nature of the survey and the long-standing relationship the parents had with the OCHL research team.

Selected demographic data from the parents and hearing measures of the children from the OCHL study were used to test for bias created by those that responded (respondents) to the survey compared to those who did not respond (nonrespondents). There was no significant difference between respondents and nonrespondents with regard to the children's better ear pure-tone average (BEPTA), age at which the child was fitted with a hearing aid, age of the child at the time of survey request, or the mother's age. Table 1 provides a summary of these comparisons. Additional analyses using chi-square tests compared respondents to nonrespondents by mother's education and income level. Mother's education was converted to four levels, which were (a) high school or less, (b) postsecondary education, (c) college graduate, and (d) postgraduate work. Income levels were converted to six levels, which were (1) < \$20,000, (2) \$20,001–\$40,000, (3) \$40,001–\$60,000, (4) \$60,001–\$80,000, (5) \$80,000–\$100,000, and (6) > \$100,000. There was a statistically significant difference in mother's education level (p = .0004) and income level (p = .0162) between respondents and nonrespondents. Table 2 shows that the nonrespondent group tended to have lower levels of maternal education and income compared to the respondents.

Table 2. Gender, education, and income distribution of respondents and nonrespondents.

Demographic and background characteristics	Respondents *n*	Respondents %	Nonrespondents *n*	Nonrespondents %
Highest educational level completed				
Completed high school or less	22	11.3	21	19.3
Postsecondary education	50	25.8	45	41.3
College graduate	55	28.4	24	22.0
Postgraduate work	63	32.5	16	14.7
Undisclosed	4	2.1	3	2.8
Household income level				
< \$20,000	12	6.2	15	13.8
\$20,001–\$40,000	18	9.3	15	13.8
\$40,001–\$60,000	40	20.6	24	22.0
\$60,001–\$80,000	39	20.1	17	15.6
\$80,001–\$100,000	27	13.9	11	10.1
> \$100,000	48	24.7	12	11.0
Undisclosed	10	5.2	15	13.8
Child's gender				
Male	102	52.6	61	56.0
Female	92	47.4	48	44.0

It is important to note that all but five of the children who had a survey returned were fitted with hearing aids. Those that were not fitted had mild hearing loss. There were also eight children who were implanted with cochlear implants over the course of the OCHL longitudinal study. All eight of these children were previously fitted with hearing aids.

Fluency Survey

The primary purpose of the fluency survey was to gather information from parents about their children's history of stuttering. The survey targeted parents because they likely had the most consistent exposure to the child during the preschool years when stuttering is most prevalent. If the parents indicated that their child had exhibited stuttering behaviors at one point in time, they were asked a series of questions regarding the nature of the stuttering behavior, the age of stuttering onset, age when the stuttering stopped (if it had), and whether the child ever received therapy for stuttering.

Table 1. Characteristics of the children and mothers of the respondents and nonrespondents of the survey.

Variable	Respondents Mean	Respondents *SD*	Nonrespondents Mean	Nonrespondents *SD*	*p* Value
Better ear PTA	48.8	13.9	49.1	14.6	0.86
Age aid fitted	15.4	17.4	17.0	18.4	0.48
Child's age at time of survey submission	8	2.1	8.4	2.1	0.22
Mother's age at time of survey submission	33.6	5.1	32.6	6.1	0.15

Note. PTA = pure-tone average at 500, 1000, 2000, and 4000 Hz.

The clinical distinction between typical disfluencies (interjections, revisions, and phrase repetitions) and stuttering-like disfluencies (monosyllabic whole-word repetitions, sound or syllable repetitions, and blocks or prolongations) may not be known to most parents, so a description of these behaviors and the distinction between typical and stuttering-like disfluencies were explicitly provided at the beginning of the survey. Although monosyllabic whole-word repetitions (e.g., *I-I-I-I* like cookies.) are typically considered to be stuttering-like disfluencies (Ambrose & Yairi, 1999) and are a common characteristic of preschool stuttering, we did not include these in the definition of stuttering provided to the parents because we did not want to confuse parents with the distinction between monosyllabic whole-word repetitions (which are considered stuttering-like repetitions) and multisyllabic whole-word repetitions (which are considered typical disfluencies). Brocklehurst (2013) argued that incidence and prevalence studies may produce inflated rates because of how stuttering is defined, in particular because of the inclusion of monosyllabic whole-word repetitions. Yairi (2013) strongly defended the inclusion of monosyllabic whole-word repetitions in the definition of stuttering, a viewpoint that we agree with. However, due to the survey's retrospective reporting of stuttering, the decision not to include whole-word repetitions in the stuttering definition aimed to reduce reports of stuttering that were possibly questionable. The Appendix includes the survey questions. The online and paper copy of the fluency survey had identical questions.

Speech, Language, and Hearing Data

Speech, language, and hearing data were collected annually from children during their participation in the OCHL longitudinal study (see Tomblin, Walker, et al., 2015, for a full description of the study design, protocols, and outcome measures). The OCHL study used an accelerated longitudinal design in which children were recruited at various ages from birth to 7 years and followed for at least 3 years. Due to the staggered age of initial participation in the study, not all children have data at all ages, and different speech and language measures were collected at each age. However, hearing data were gathered for all children at all visits. The two hearing measures used for group comparisons were BEPTA and aided speech intelligibility index (SII; American National Standards Institute, 1997). BEPTA is a means for describing the degree of hearing loss; it was calculated by taking the average of unaided hearing thresholds for pure-tone stimuli at three or four frequencies in the better ear. BEPTA of less than 20 dB HL represents hearing within normal limits; BEPTA between 20 and 40 dB HL represents mild hearing loss; BEPTA between 41 and 60 dB HL represents moderate hearing loss; and BEPTA between 61 and 80 dB HL represents severe hearing loss. Rather than describing access to sound in terms of unaided thresholds to pure-tone stimuli, aided audibility, quantified by the SII, was used. Aided SII is a means for describing the percentage of the long-term average speech spectrum that is accessible with hearing aids at a given input level. SII is measured on a scale of 0–1, with 0 representing *no access to the speech spectrum* and 1 representing *full access to the speech spectrum*. It more accurately represents the everyday listening experiences of CHH than unaided pure-tone average. It can be used to account for amplification characteristics of hearing aids, including proximity of hearing aid fit to prescriptive targets (McCreery et al., 2015), variability in ear canal acoustics, and amount of access to the speech spectrum at a conversational level (Stiles, Bentler, & McGregor, 2012).

Speech and language analyses in this study focused on the 3- and 4-year visits to compare speech, language, and hearing data at the ages when stuttering tends to first emerge (the average age of stuttering onset in this sample was 3.71 years old). The 3-year speech and language data consisted of the standard scores from the Goldman-Fristoe Test of Articulation–Second Edition (GFTA-2; Goldman & Fristoe, 2000), Vineland Adaptive Behavior Scales–Second Edition (Vineland-II; Sparrow et al., 2005) composite scores, the Comprehensive Assessment of Spoken Language (CASL; Carrow-Woolfolk, 1999) composite score, and scaled scores from subtests of the MacArthur–Bates Communicative Development Inventory Upper Extension (MBCDI; Fenson et al., 1993). The 4-year speech and language data consisted of the standard scores from the Vineland and CASL composite scores, Test of Preschool Early Literacy (TOPEL; Lonigan, Wagner, Torgesen, & Rashotte, 2007) phonological awareness subtest, scaled scores from selected Vineland-II subtests, and the Wechsler Preschool and Primary Scale of Intelligence–Third Edition (WPPSI-III; Wechsler, 2002) vocabulary subtest.

Data Analysis

Most of the data from the survey were categorical. Exact binomial tests were conducted on the incidence and prevalence data to compare our sample to the theoretically expected distribution obtained from population estimates in the literature. The exact binomial test is used to test the null hypothesis that two categories are equally likely to occur and does not rely on an asymptotic normality assumption. To relate continuous variables such as age of stuttering onset and speech, language, and hearing variables from OCHL to stuttering status, we used two-sample t tests with a Satterthwaite adjustment for unequal variances. In addition, we used a stepwise selection procedure with logistic regression to determine which of the variables were the most important in determining stuttering status. Specifically, the outcome variables in the models were children who have exhibited stuttering versus those who never stuttered, and children who were still stuttering at the time of survey submission compared to children who had stopped stuttering. All significance tests were evaluated at the standard .05 level of significance. Due to the exploratory nature of this study, no alpha adjustments were made for multiple tests. Where appropriate, Cohen's d effect sizes are reported. Analyses were carried out in SPSS (Version 23;

IBM Corp., 2015) and R package (Version 3.3.2; R Core Team, 2016).

Results

A summary and descriptive statistics of the responses to the fluency survey are presented in Table 3. The individual responses to the survey from those who stated that their child had stuttered at one point in time are provided in the Supplemental Material.

Research Question 1: What is the Incidence and Prevalence of Stuttering in CHH?

From the 194 surveys returned, 33 children were identified as having exhibited stuttering behaviors at some point in their life, resulting in an incidence of 17.01%. An exact binomial test comparing the incidence from this sample to an estimated incidence of 8% in the general population (Yairi & Ambrose, 2013) yielded a significant difference ($p < .0001$, 95% CI [0.120, 0.231]). Ten children were stuttering at the time of survey submission, corresponding with a prevalence of 5.15%. An exact binomial test, comparing the prevalence from this sample to an estimated prevalence of 1.5% in the general population in the age group of most of the children who were surveyed, yielded a significant difference ($p = .0008$, 95% CI [0.025, 0.093]). The 1.5% estimated prevalence used for this analysis is a conservative estimate for school-age children based on the synthesis of prevalence studies from Yairi and Ambrose (2013).

Research Question 2: What Are the Characteristics of Stuttering in CHH?

Age of Onset

The average age of stuttering onset for all the children who were reported to have stuttered was 3.7 years ($SD = 1.4$). The average age of onset for the children who had stopped stuttering (from this point on, to be referred to as recovered) was 3.2 years ($SD = 1.1$). The average age of onset for the children who were still stuttering at the time of survey submission was 4.8 years ($SD = 1.7$). A two-sample t test, with a Satterthwaite adjustment for unequal group variance, showed a significant difference in the age of onset of those who have recovered and those who were still stuttering at the time of the survey ($p = .02$).

Age of Recovery and Length of Time Stuttering Before Recovery

For the children who recovered, the average age at which they stopped stuttering was 4.59 years ($SD = 1.16$). However, the average age of recovery was based on 16 children, because data were not provided about the age stuttering stopped for seven of the 23 children who recovered. The average length of time stuttering before recovery was calculated by subtracting the age when stuttering stopped from the age of onset, which yielded an average of 1.52 years ($SD = 1.33$).

Types of Disfluencies

Written descriptions of the types of stuttering that the child exhibited were provided by the survey respondents for all of the children who had stuttered. These descriptions are presented in the Supplemental Material. The descriptions

Table 3. Age, therapy, secondary, and avoidance behavior data from the survey.

Variable	Frequency	%	Mean	*SD*
Age at survey submission	194	100	8.0	2.1
Never stuttered	161	83.0	8.1	2.1
Male	79	49.1	8.1	2.2
Female	82	51.0	8.2	2.1
Stuttered at some point (incidence)	33	17.0	7.6	1.8
Male	23	69.7	7.7	1.7
Female	10	30.3	7.3	1.9
Still stuttering at time of survey (prevalence)	10	5.15	7.3	1.4
Male	6	60	7.2	1.8
Female	4	40	7.4	0.9
Age of stuttering onset	33	100	3.7	1.4
No longer stuttering	23	69.7	3.2	1.1
Still stuttering at time of survey	10	30.3	4.8	1.7
Age stuttering stopped in the recovered group[a]			4.6	1.2
Exhibited secondary behaviors ($n = 33$)	6	18.2		
No longer stuttering ($n = 23$)	3	13		
Still stuttering at time of survey ($n = 10$)	3	30		
Exhibited avoidance behaviors ($n = 33$)	8	24.2		
No longer stuttering ($n = 23$)	5	21.7		
Still stuttering at time of survey ($n = 10$)	3	30		
Received therapy for stuttering ($n = 33$)	12	36.4		
No longer stuttering ($n = 23$)	8	34.8		
Still stuttering at time of survey ($n = 10$)	4	40		

[a]No age of when stuttering stopped was provided for seven of the 23 children who had recovered.

were coded by disfluency type based on the way in which stuttering behaviors were described at the beginning of the fluency survey (repetitions, prolongations, and blocks). Table 4 shows the frequency counts for each type of disfluency that was described, as well as mutually exclusive categories of the manner in which the children stuttered. Repetitions were the most common type of stuttering, with 72.7% of children demonstrating this type of disfluency. These repetitions should be interpreted to be sound or syllable repetitions given that the description of disfluencies at the beginning of the survey explicitly stated that whole-word repetitions were not stuttering-like behaviors. As stated before, we are not stating that monosyllabic whole-word repetitions are not stuttering-like disfluencies; however, to be conservative, we decided not to include whole-word repetitions in this study. Prolongations were the next most common type of disfluency (39.4%), followed by blocks (27.3%).

Mutually exclusive categories of stuttering descriptions showed that close to half of the children only exhibited repetitions (42.4%), with a much smaller percentage only exhibiting prolongations (21.2%) or only blocks (6.1%). The rest of the children had repetitions and some combination of other stuttering-like disfluencies.

Secondary Behaviors

Nine of the children were reported to display only secondary behaviors. Four responses described the secondary behaviors, which are provided in the Supplemental Material. Some examples of secondary behaviors are “failure to make eye contact,” “shows signs of tensing up and straining,” and “eye blinking, sniffing.”

Avoidance Behaviors

Eleven children showed avoidance behaviors. Three children had both secondary behaviors and avoidance behaviors. Eight survey responses described the avoidance behaviors, which are included in the Supplemental Material. The most common theme regarding avoidance was choosing not to speak or avoiding social situations (e.g., “she will choose not to talk and instead try to avoid finishing the sentence or statement or say never mind” and “avoiding socializing with kids”).

Table 4. Types of disfluent behavior reported in the survey.

Disfluencies	*n*	%
Types of disfluencies described		
Repetitions	24	72.7
Prolongations	13	39.4
Blocks	9	27.3
Mutually exclusive categories of disfluent behavior		
Only repetitions	14	42.4
Only prolongations	7	21.2
Only blocks	2	6.1
Repetitions and prolongations	3	9.1
Repetitions and blocks	4	12.1
Repetitions, prolongations, and blocks	3	9.1

Treatment for Stuttering

Twelve of the children were reported to have received therapy. Descriptions regarding the details of the therapy are presented in the Supplemental Material. Several of the descriptions were rather vague (e.g., “speech,” “speech therapy,” “speech therapy birth to present,” and “he was already receiving speech therapy, stuttering only lasted couple of weeks and stopped”), making it difficult to know whether they were indicating that the child had received speech therapy in general or whether therapy specifically targeted stuttering. Of the 10 children who were still stuttering at the time of the survey, four were indicated to have received therapy for stuttering, but descriptions were unclear whether therapy was currently occurring and whether the therapy specifically targeted stuttering. One of the therapy descriptions for a child who was still stuttering indicated “they are aware of it at speech, but are choosing not to treat it at this time.”

Research Question 3: In CHH, Around the Average Age of Stuttering Onset, Are There Differences in the Speech, Language, or Hearing Abilities Between Children With and Without a History of Stuttering, and Are There Differences Between Children Who Recovered From Stuttering Versus Those Who Are at Risk of Persisting?

CHH Who Stuttered Versus Those Who Never Stuttered

Speech, language, and hearing data obtained from the OCHL dataset were used to compare children who were reported to have stuttered to children who never stuttered. Table 5 shows descriptive data for each variable using the 3- and 4-year OCHL data. For audiological measures, no significant difference was found in BEPTA, but there was a significant difference in SII where children who never stuttered had lower audibility (M = 0.81, SD = 0.10, for children who stuttered; M = 0.76, SD = 0.14, for children who never stuttered; p = .032, d = 0.411). In the 3-year comparison, all of the speech and language variables except MBCDI vocabulary, Vineland receptive, and CASL composite were significant at the .05 level. Several variables had medium effect sizes: GFTA (d = 0.595), MBCDI language (d = 0.702), Vineland expressive language (d = 0.638), Vineland socialization composite (d = 0.530), Vineland motor composite (d = 0.659), and Vineland adaptive behavior composite (d = 0.751). The MBCDI sentence had a large effect size (d = 0.811). We know that many of these variables are highly correlated with each other, so we used a stepwise logistic regression analysis to determine which variables contribute the most unique information toward stuttering status. Due to missing data at the 3-year visit for some of the children, the logistic regression at this age included 14 children who had stuttered and 40 who never stuttered. In the stepwise logistic regression, Vineland

Table 5. Three- and 4-year data of children who have and have not stuttered.

Variable	Stuttered at some point			Never stuttered				
	n	Mean	*SD*	*n*	Mean	*SD*	*p*	*d*
Better ear pure-tone average	33	46.2	11.6	161	49.3	14.3	.25	0.24
Better ear speech intelligibility index	33	0.81	0.1	159	0.76	0.14	.03	0.41
3-year data								
GFTA	18	79.1	14.2	62	89.4	20.0	.02	0.59
MBCDI language	17	8.2	6.6	47	21.3	25.4	.00	0.70
MBCDI vocabulary	17	12.6	20.4	47	21.0	25.9	.19	0.36
MBCDI sentence	17	9.7	6.5	47	18.8	14.5	.00	0.81
Vineland receptive language	20	13.5	3.1	58	14.8	3.1	.10	0.43
Vineland expressive language	20	12.6	2.0	58	14.2	3.0	.01	0.64
Vineland communication composite	20	87.0	13.6	58	94.7	18.0	.05	0.48
Vineland socialization composite	20	90.3	8.9	58	96.2	13.1	.03	0.53
Vineland motor composite	20	88.2	11.7	58	96.2	12.7	.01	0.66
Vineland adaptive behavior composite	20	87.9	9.6	58	96.2	12.5	.00	0.75
CASL composite	19	82.7	15.1	59	90.8	17.8	.06	0.49
4-year data								
Vineland receptive language	20	14.7	2.7	77	14.6	3.0	.89	0.03
Vineland expressive language	20	13.3	2.9	77	14.1	2.7	.26	0.30
Vineland communication composite	20	96.7	11.8	77	97.8	12.9	.72	0.09
Vineland socialization composite	20	95.5	9.8	77	98.3	13.6	.31	0.23
Vineland motor composite	20	91.8	13.8	77	97.2	15.0	.14	0.37
Vineland adaptive behavior composite	20	93.6	11.7	77	97.7	13.3	.19	0.33
CASL composite	21	94.3	16.9	78	98.9	20.5	.30	0.24
TOPEL phonological awareness	20	90.3	17.9	72	93.8	16.7	.44	0.20
WPPSI vocabulary	18	8.6	2.4	70	8.7	3.4	.80	0.06

Note. GFTA = Goldman-Fristoe Test of Articulation; MBCDI = MacArthur–Bates Communicative Development Inventory; CASL = Comprehensive Assessment of Spoken Language; TOPEL = Test of Preschool Early Literacy; WPPSI = Wechsler Preschool and Primary Scale of Intelligence.

expressive was entered into the model as the most significant variable ($p < .01$), and SII was entered second ($p = .04$). None of the other variables were added, indicating that they did not add significant information into the model that was not already captured by Vineland expressive or SII. Results indicate that the odds of not stuttering was 2.07 times higher for a 1-unit higher Vineland expressive score. The logistic regression analysis with the 4-year data included 17 who stuttered and 61 who never stuttered. No variable reached statistical significance.

CHH Who Recovered From Stuttering Versus Those Who Are at Risk of Persisting

This analysis is particularly exploratory given that there is a high likelihood that some of the children who were still stuttering at the time the survey was submitted will end up recovering. This was especially true for two of the children who had been stuttering for less than 2 years and were under 6 years of age. To be more conservative, children in the "at risk of persistence" group were included only if they had been stuttering for more than 24 months or they were over 8 years old. These were deemed to be reasonable cutoff criteria given that the children needed to either have a late onset of stuttering or a long history of stuttering, both of which are risk factors for persistence (Yairi & Ambrose, 2005). Using these criteria, there were eight children who were at risk of persistence and had OCHL outcome data at either the 3- or 4-year visit. Speech, language, and hearing data from the "at risk of persistence" group were compared to the children who had stuttered at one point but had recovered. A summary of the outcome comparisons are presented in Table 6.

We conducted *t* tests to determine whether statistically significant differences existed between groups. Medium or greater effect sizes are reported. There was a significant difference and large effect size between age of onset between the recovered ($M = 3.2$, $SD = 1.1$) and the "at risk of persistence" group ($M = 4.8$, $SD = 1.9$, $p = .05$, $d = 1.03$). There was also a statistically significant difference and large effect size in TOPEL phonological awareness for recovered ($M = 94.2$, $SD = 19.8$) versus still stuttering ($M = 81.2$, $SD = 7.2$, $p = .045$, $d = 0.875$), with the recovered group having higher scores. Although not statistically significant, based on standard rules for assessing effect sizes (Sawilowsky, 2009), there was a medium effect size in better ear SII with the recovered group ($M = 0.82$, $SD = 0.11$) having greater audibility than the group who still stutter ($M = 0.77$, $SD = 0.07$, $p = .169$, $d = 0.542$). There was a medium effect size difference on the MBCDI language composite at the 3-year visit with children in the recovered group ($M = 8.9$, $SD = 7.8$), outperforming children in the "at risk of persistence" group ($M = 5.8$, $SD = 2.0$, $p = .291$, $d = 0.534$). There was also a medium effect size difference on the CASL composite at the 3-year visit with children in the recovered group ($M = 86.1$, $SD = 16.9$), outperforming children in the "at risk of persistence" group ($M = 77.7$, $SD = 14.0$, $p = .299$, $d = 0.541$). A similar trend in group comparison existed with a large effect size difference on WPPSI-III vocabulary ($M = 9.2$, $SD = 2.5$,

Table 6. Three- and 4-year data of children at risk of persistence and those who recovered.

	At risk of persisting			Recovered				
Variable	*n*	Mean	*SD*	*n*	Mean	*SD*	*p*	*d*
Better ear pure-tone average	8	50.2	12.7	23	44.8	11.6	.31	0.44
Better ear speech intelligibility index	8	0.77	0.07	23	0.82	0.11	.17	0.54
3-year data								
GFTA	5	76.6	8.0	11	78.8	17.2	.73	0.17
MBCDI language	6	5.8	2.0	9	8.9	7.8	.29	0.53
MBCDI vocabulary	6	8.3	6.1	9	7.2	3.6	.70	0.22
MBCDI sentence	6	9.2	3.8	9	9.4	8.5	.93	0.04
Vineland receptive language	7	12.7	3.0	11	13.6	3.3	.56	0.29
Vineland expressive language	7	12.4	0.8	11	12.5	2.7	.89	0.06
Vineland communication composite	7	85.1	11.1	11	87.5	15.9	.72	0.17
Vineland socialization composite	7	88.6	7.4	11	89.0	9.0	.91	0.05
Vineland motor composite	7	83.9	10.7	11	89.4	12.7	.34	0.47
Vineland adaptive behavior composite	7	84.7	7.1	11	87.9	10.6	.46	0.35
CASL composite	7	77.7	14.0	10	86.1	16.9	.28	0.54
4-year data								
Vineland receptive language	7	14.4	3.7	13	14.8	2.0	.83	0.11
Vineland expressive language	7	13.3	2.3	13	13.3	3.3	.99	0.01
Vineland communication composite	7	97.1	12.2	13	96.5	12.0	.91	0.06
Vineland socialization composite	7	97.6	12.7	13	94.4	8.2	.56	0.30
Vineland motor composite	7	93.7	13.8	13	90.8	14.2	.66	0.21
Vineland adaptive behavior composite	7	96.0	14.2	13	92.3	10.5	.56	0.30
CASL composite	7	89.9	8.4	14	96.6	19.8	.29	0.44
TOPEL phonological awareness	6	81.2	7.2	14	94.2	19.8	.05	0.88
WPPSI vocabulary	6	7.3	1.5	12	9.2	2.5	.07	0.88

Note. GFTA = Goldman-Fristoe Test of Articulation; MBCDI = MacArthur–Bates Communicative Development Inventory; CASL = Comprehensive Assessment of Spoken Language; TOPEL = Test of Preschool Early Literacy; WPPSI = Wechsler Preschool and Primary Scale of Intelligence.

for children who recovered; $M = 7.3$, $SD = 1.5$, for children in the "at risk of persistence" group; $p = .123$, $d = 0.884$) at the 4-year visit.

It is important to note that the only child reported to have stuttered who also received cochlear implant is one of the children in the "at risk of persistence" group. This child was implanted at 2.12 years of age and began stuttering at age 3; at the time of the survey, the child was 7.91 years old. To our knowledge, this is the first report of a child with a cochlear implant who seems likely to persist in stuttering.

Discussion

This study used a parent questionnaire to investigate the characteristics of stuttering in CHH. The questionnaire included information regarding age of stuttering onset, age of recovery, types of disfluencies, secondary and avoidance behaviors, and treatment of stuttering. Speech, hearing, and language data from the OCHL study were used to compare CHH who never stuttered to CHH who did stutter at some point. We also compared children who were "at risk of persistence" to children who recovered from stuttering.

Incidence and Prevalence

Both the incidence and prevalence of stuttering were higher in this sample of CHH compared to estimates of the general public. The incidence in this sample was 17.1%, significantly greater than an estimated population incidence of 8% (Yairi & Ambrose, 2013). The prevalence in this sample was 5.15%, which was greater than an estimated prevalence of 1.5% in the general population in the age range of most of the children who were surveyed (Yairi & Ambrose, 2013). These findings are important given the previous research showing a lower prevalence of stuttering in children with hearing loss (Backus, 1938; Harms & Malone, 1939; Montgomery & Fitch, 1988). There are several reasons why our findings may contradict previous studies. First, previous studies targeted school-age children so they would not have detected children who recovered from stuttering. Second, the earlier surveys were only sent to teachers in schools for the deaf. These schools likely had a disproportionately high level of students with severe to profound hearing loss, and there was little information regarding the children's use of speech versus sign. Third, the previous studies are relatively old, with two of the largest studies conducted in the 1930s and a third study conducted in the mid-1980s. The two oldest studies likely had children with limited access to amplification due to late identification of hearing loss and use of less sophisticated technologies than currently available. Even in the 1980s, the sophistication and use of hearing aids were limited compared to the recent advances in technology and early identification. The higher incidence and prevalence results in the current study were likely influenced by the fact that we specifically targeted children who had a mild to moderately severe hearing loss, they all communicated via spoken language, and most received hearing aids at a relatively early age. Theoretical explanations for why CHH may be more susceptible to stuttering are discussed later.

Characteristics of CHH Who Stutter and Therapy Received

The characteristics of stuttering and its onset and recovery in this sample of CHH are very similar to hearing children who stutter. The age of onset (M = 3.7 years), length of stuttering before recovery (M = 1.5 years), age of recovery (M = 4.6 years), types of disfluencies, and descriptions of secondary behaviors are all consistent with what has been described in longitudinal studies of hearing children who stutter (Yairi & Ambrose, 1999). The similarity of these characteristics between CHH and hearing children leads to the conclusion that early intervention of stuttering should be introduced and administered in similar proportions across both these populations.

Despite the high incidence of stuttering in this sample of CHH and the reports of struggle and avoidance behaviors, there was a striking lack of stuttering intervention provided to these children. Of the 10 children who were still stuttering at the time of the survey, four received therapy according to parent report. All descriptions of therapy were rather unspecific as to whether therapy was currently occurring and whether the therapy specifically targeted stuttering. For example, in one case a parent reported, "The hearing impaired teacher in the More at Four program is working with him." In this case, it was not clear based on this description whether the teacher for the deaf/hard of hearing was planning the stuttering therapy or whether they were implementing strategies within the classroom to facilitate fluency as instructed by a speech-language pathologist. One of the therapy descriptions for a child who was still stuttering indicated "they are aware of it at speech, but are choosing not to treat it at this time." This highlights some of the ambiguity of the treatment descriptions: Either the child was receiving stuttering therapy previously but it was discontinued or the parent's "yes" response to stuttering therapy was referring to speech therapy in general.

Given that most early intervention programs for stuttering recommend some form of intervention (whether direct or indirect) if stuttering has not begun to resolve within 18 months, all of the eight children in the "at risk of persistence" group should be receiving some form of stuttering intervention. However, only one of those eight indicated that they are receiving stuttering intervention. The other seven children all exhibited, based on parents' survey responses, well-established risk factors for persistence based on their current age and length of time since stuttering onset. Several of them also were reported to have secondary and/or avoidance behaviors and had descriptions like "frustration, failure to make eye contact" and "she will choose not to talk and instead try to avoid finishing the sentence or statement or say never mind." Due to the survey nature of this study, it cannot be determined what role hearing loss may be playing in avoidance behaviors. Perhaps the most striking example where therapy seemed to be warranted was with a child where therapy was described as "they are aware of it at speech, but are choosing not to treat it at this time." At the time of the survey, this child was 8 years old and had been stuttering since age 5, and the parent reported "repetitions of the beginnings of a word, prolongations, and blocks" and avoidance behaviors described as "tried to avoid saying certain words."

Differences Between CHH Who Have Never Stuttered to Those Who Have Stuttered

Across a range of tests, children who never stuttered displayed significantly greater speech and language skills at 3 years of age compared to children who stuttered at some point. These results fit with findings from a meta-analysis, which showed that there are subtle but significant differences between the overall language abilities of hearing children who stutter compared to those who do not stutter (Ntourou et al., 2011). If language proficiency is a contributing factor for stuttering onset (Conture & Walden, 2012; Smith & Weber, 2016), then the language delays seen in CHH (Tomblin, Harrison, et al., 2015) may be contributing to the higher incidence of stuttering in this sample. Furthermore, there was a significant group difference in the 3-year Vineland expressive, but not the Vineland receptive, demonstrating a potential disassociation, something that has been hypothesized to be a risk factor for stuttering (Anderson et al., 2005; Clark et al., 2015; Coulter et al., 2009). However, there were no significant differences in speech or language measures at 4 years of age between children who never stuttered and children who stuttered. Given that the average age of stuttering onset was 3.7 years, language delays or disassociation between expressive and receptive language may precede stuttering onset but dissipate shortly after. There was also a significant difference, with a small effect size, in aided SII: Children who stuttered at some point in time had higher aided audibility than the CHH who never stuttered. These results are discussed in more detail in the next section.

Differences Between CHH Who Have Recovered From Stuttering and Those Who Are at Risk of Persistence

Exploratory analyses investigated variables that differentiated CHH who recovered from stuttering (n = 20) from CHH who were still stuttering at the time of the survey and who showed significance risk factors for persistence (n = 8). There was a significant difference (with a large effect size) between the groups on the TOPEL phonological awareness at age 4. Phonological awareness is a measure of a child's understanding that speech is made up of abstract units (e.g., syllables, onset and rime units, and individual phonemes). This result is consistent with that of Paden, Yairi, and Ambrose (1999), who found that children who persisted in stuttering had poorer phonological abilities compared to children who recovered from stuttering when assessed close to stuttering onset. A second significant difference (with a large effect size) between the two groups was that the "at risk of persistence" group had a later average age of onset. This finding is consistent with longitudinal studies, which showed that older age of onset is a risk factor for persistence (Yairi & Ambrose, 2005). Due to the small sample size in these analyses, there were several medium to large effect size differences that did not reach statistical significance. For example, there were medium effect sizes where the recovered group outperformed the "at risk of persistence" group on the MBCDI language composite and the CASL composite and had higher SII scores. There was also a similar group difference, but with a large effect size on the WPPSI-III vocabulary. Collectively, between the differences in age of onset, phonological awareness, and other hearing and language variables with medium to large effect sizes, the factors that may be related to persistent stuttering in CHH are similar to variables that have been identified as risk factors in hearing children (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Paden et al., 2002; Yairi & Ambrose, 2005; see Smith & Weber, 2016, for a synopsis of the longitudinal work of the Purdue Stuttering Project). It needs to be emphasized that these were exploratory analyses with a small sample size, so further research is warranted.

If indeed the recovered group has greater audibility than the "at risk of persistence" group, as measured by SII, it may have theoretical implications regarding the relationship between audibility and stuttering. SII is a means of describing the percentage of the long-term average speech spectrum that is accessible with hearing aids at a given input level (e.g., 65 dB SPL or conversational speech). It more accurately represents the everyday listening experiences of CHH than unaided pure-tone average (McCreery et al., 2015; Stiles et al., 2012). With the substantial evidence that audition and auditory motor integration are involved in fluency, poorer audibility could be a risk factor for persistent stuttering. According to hypotheses of stuttering behavior based on the Directions Into Velocities of Articulators model, disfluencies occur when a sufficient degree of mismatch between feedforward motor commands and perceptual feedback triggers a reset or a change in motor behavior that is observed as stuttering (Civier et al., 2010; Max, Guenther, Gracco, Ghosh, & Wallace, 2004). Although explanations of stuttering using this model have mostly implicated deficits in the feedforward system and an overreliance on feedback, it is also feasible that similar mismatches between feedforward and feedback could occur if the auditory targets are less developed. Less defined auditory targets may lead to less accurate auditory perception and decreased feedforward stability, which could ultimately lead to larger mismatches during speech production and an increased risk of disfluencies.

Howell, Davis, and Williams (2006) showed that hearing children who stutter have decreased auditory abilities compared to hearing children who do not stutter, and auditory abilities differentiate children who recovered from stuttering from those who persisted. Howell, Rosen, Hannigan, and Rustin (2000) demonstrated that a group of children who stutter had poorer central auditory processing abilities, as measured by backward masking thresholds, compared to fluent age-matched children. The backward masking thresholds were also correlated with stuttering severity. Howell et al. (2006) found that backward masking thresholds were significantly different between adolescents who stutter compared to those who had recovered from stuttering. This provides evidence that precise auditory perception may play a role in fluent speech production. Lower audibility may put children at greater risk of stuttering persistence due to less quality auditory speech input that is necessary to develop the integration of refined auditory targets in combination with an adequate feedforward speech motor system. However, the children who stuttered in this study had greater audibility than children who never stuttered, demonstrating that audibility may be more related to persistence or recovery and not stuttering onset.

Clinical Implications

The current study shows that the characteristics of stuttering and potential risk factors for persistence may be similar in CHH compared to hearing children. However, on the basis of this sample, CHH may have a higher incidence and prevalence of stuttering. Increased stuttering may be related to the speech and language challenges that CHH face. Despite the increased incidence of stuttering, this study showed low degree of stuttering intervention in CHH. This may be related to the fact that CHH often have significant speech and language delays (Tomblin, Harrison, et al., 2015) that are being addressed instead of the stuttering. Given the psychosocial repercussions such as being bullied, increased risk of social anxiety, decreased vocational opportunities, and decreased overall life satisfaction that are associated with chronic stuttering (Blood, Blood, Maloney, Meyer, & Qualls, 2007; Blumgart, Tran, & Craig, 2010), it is important that speech-language pathologists are well informed of the risks and negative outcomes of stuttering. If in fact there is a lack of stuttering intervention in CHH, as the current study suggests, there needs to be an education campaign aimed at speech-language pathologists who commonly work with CHH to increase knowledge and clinical preparedness to address stuttering.

Limitations and Future Directions

This investigation had several limitations. First, survey data have inherent limitations compared to direct observations or interviews. For example, several descriptions of stuttering treatment lacked clarity and specificity, making it difficult to draw clear conclusions about the nature of treatment received. Second, there was a limitation in the retrospective nature of the survey and a lack of direct assessment of stuttering behaviors. The average age of the children at the time of survey submission was 7.4 years old, so the parents were asked to remember speech patterns that may have occurred several years in the past. It could be argued that parent reports are not as reliable as direct observation by a fluency specialist. However, there is evidence that there is strong agreement between parent reports and clinician identification of stuttering (Curlee, 2007; Yairi & Ambrose, 2005). To address the potential limitations of retrospective parental reports, the definition of stuttering behavior provided to the parents was limited to sound/syllable repetitions and audible/inaudible prolongations, excluding monosyllabic whole-word repetitions that most researchers categorize as stuttering-like disfluencies (Yairi, 2013). This conservative definition may have actually biased the results toward underreporting of stuttering. Third, only a subset of the children had complete speech and language datasets from the OCHL study, so the relationships between speech and language factors and stuttering should be interpreted with caution.

Future studies should further investigate the prevalence of stuttering in preschools that specialize in early intervention and education for children with hearing loss to allow for direct observation of stuttering behaviors in this population. With the increased enrollment in early intervention for CHH, a survey of school-based speech-language pathologists would be important to understand the views of stuttering in this population, as well as the likelihood of stuttering intervention with CHH. It would also be beneficial to develop best practice recommendations for how to integrate stuttering intervention with the other speech/language services that are most often provided to CHH. Such recommendations would increase education of stuttering for speech-language pathologists who focus on working with children with hearing loss, hopefully leading to increased stuttering intervention in this population.

Another possible future direction is investigating the phenomena that influence stuttering frequency in children with cochlear implants. Due to the nature of the technology, this population may provide a unique opportunity to study the role of audition in stuttering. For example, if a child stutters and has a cochlear implant, it would be informative to test what happens to their fluency if the implants were temporarily turned off. Such a reduction in audition and hence auditory feedback is difficult in hearing children without high levels of masking. In the case of cochlear implants, it may be possible to completely, or nearly completely, eliminate auditory feedback without the use of masking, which itself may introduce changes to the auditory motor integration during speech.

Conclusion

The current results suggest that the incidence and prevalence of stuttering in children with mild to moderately severe hearing loss is as high, if not higher, than hearing children. Given the delays in speech and language that this population exhibits, reduced auditory access may put these children at higher risk for both the emergence and persistence of stuttering. Clinicians who work with CHH should be aware of this potential increased risk and plan interventions that address stuttering in addition to any other communication needs.

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References

Ambrose, N. G., & Yairi, E. (1999). Normative disfluency data for early childhood stuttering. *Journal of Speech, Language, and Hearing Research, 42*(4), 895–909. https://doi.org/10.1044/jslhr.4204.895

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

American National Standards Institute. (1997). *American national standards methods for the calculation of the articulation index* (ANSI S3.5-1997). New York, NY: Author.

Anderson, J. D., Pellowski, M. W., & Conture, E. G. (2005). Childhood stuttering and dissociations across linguistic domains. *Journal of Fluency Disorders, 30*(3), 219–253. https://doi.org/10.1016/j.jfluids. 2005.05.006

Andrews, G., Craig, A., Feyer, A., Hoddinott, S., Howie, P., & Neilson, M. (1983). Stuttering: A review of research findings and theories circa 1982. *Journal of Speech and Hearing Disorders, 48,* 226–246.

Arndt, J., & Healey, E. C. (2001). Concomitant disorders in school-age children who stutter. *Language, Speech, and Hearing Services in Schools, 32,* 68–78.

Backus, O. (1938). Incidence of stuttering among the deaf. *Annals of Otology, Rhinology, and Laryngology, 47,* 632–635.

Blood, G. W., Blood, I. M., Maloney, K., Meyer, C., & Qualls, C. D. (2007). Anxiety levels in adolescents who stutter. *Journal of Communication Disorders, 40,* 452–469.

Blood, G., Ridenour, V., & Qualls, C. (2003). Co-occurring disorders in children who stutter. *Journal of Communication Disorders, 36,* 427–448.

Blood, G. W., & Seider, R. (1981). The concomitant problems of young stutterers. *Journal of Speech and Hearing Disorders, 46,* 31–33.

Bloodstein, O., & Ratner, N. (2008). *A handbook on stuttering* (6th ed.). New York, NY: Thomson Delmar Learning.

Blumgart, E., Tran, Y., & Craig, A. (2010). Social anxiety disorder in adults who stutter. *Depression and Anxiety, 27,* 687–692.

Brocklehurst, P. H. (2013). Stuttering prevalence, incidence and recovery rates depend on how we define it: Comment on Yairi & Ambrose' article Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders, 38*(3), 290–293. https://doi.org/10.1016/j.jfludis.2013.01.002

Buck, S., Lees, R., & Cook, F. (2002). The influence of family history of stuttering on the onset of stuttering in young children. *Folia Phoniatrica et Logopaedica, 54,* 117–124.

Byrd, C. T., Bedore, L. M., & Ramos, D. (2015). The disfluent speech of bilingual Spanish–English children: Considerations for differential diagnosis of stuttering. *Language, Speech, and Hearing Services in Schools, 46*(1), 30–43. https://doi.org/10.1044/2014_LSHSS-14-0010

Carrow-Woolfolk, E. (1999). *Comprehensive Assessment of Spoken Language*. Circle Pines, MN: American Guidance Service.

Ching, T. Y., Dillon, H., Marnane, V., Hou, S., Day, J., Seeto, M., . . . Zhang, V. (2013). Outcomes of early- and late-identified children at 3 years of age: Findings from a prospective population-based study. *Ear and Hearing, 34*(5), 535–552.

Civier, O., Tasko, S. M., & Guenther, F. H. (2010). Overreliance on auditory feedback may lead to sound/syllable repetitions: Simulations of stuttering and fluency-inducing conditions with a neural model of speech production. *Journal of Fluency Disorders, 35*(3), 246–279. https://doi.org/10.1016/j.jfludis.2010.05.002

Clark, C. E., Conture, E. G., Walden, T. A., & Lambert, W. E. (2015). Speech-language dissociations, distractibility, and childhood stuttering. *American Journal of Speech-Language Pathology, 24,* 480–503.

Conture, E. G., & Walden, T. A. (2012). Dual diathesis-stressor model of stuttering. In L. Bellakova & Y. Filatova (Eds.), *Theoretical issues of fluency disorders* (pp. 94–127). Moscow, Russia: National Book Centre.

Coulter, C. E., Anderson, J. D., & Conture, E. G. (2009). Childhood stuttering and dissociations across linguistic domains: A replication and extension. *Journal of Fluency Disorders, 34,* 257–278. https://doi.org/10.1016/j.jfludis.2009.10.005

Craig, A., Hancock, K., Tran, Y., Craig, M., & Peters, K. (2002). Epidemiology of stuttering in the community across the entire life span. *Journal of Speech, Language, and Hearing Research, 45,* 1097–1105.

Curlee, R. (2007). Identification and case selection guidelines for early childhood. In E. Conture & R. Curlee (Eds.), *Stuttering and related disorders of fluency* (3rd ed., pp. 3–22). New York, NY: Thieme.

Donahue, A. (2007). Guest editorial: Current state of knowledge—Outcomes research in children with mild to severe hearing loss. *Ear and Hearing, 28*(6), 713–714.

Eisenberg, L. S., Widen, J. E., Yoshinaga-Itano, C., Norton, S., Thal, D., Niparko, J. K., & Vohr, B. (2007). Current state of knowledge: Implications for developmental research—Key issues. *Ear and Hearing, 28*(6), 773–777.

Fenson, L., Dale, P. S., Reznick, J. S., Thal, D., Bates, E., Hartung, J. P., . . . Reilly, J. S. (1993). *MacArthur Communicative Development Inventories*. San Diego, CA: Singular Publishing Group.

Goldman, R., & Fristoe, M. (2000). *Goldman-Fristoe Test of Articulation–Second Edition*. Circle Pines, MN: AGS.

Harms, M., & Malone, J. (1939). The relationship of hearing acuity to stammering, *Journal of Speech and Hearing Disorders, 4,* 363–370.

Holte, L., Walker, E., Oleson, J., Spratford, M., Moeller, M. P., Roush, P., . . . Tomblin, B. (2012). Factors influencing follow-up to newborn hearing screening for infants who are hard-of-hearing. *American Journal of Audiology, 21*(2), 163–174. https://doi.org/10.1044/1059-0889(2012/12-0016)

Howell, P., Davis, S., & Williams, R. (2008). Late childhood stuttering. *Journal of Speech, Language, and Hearing Research, 51,* 669–684.

Howell, P., Davis, S., & Williams, S. M. (2006). Auditory abilities of speakers who persisted, or recovered, from stuttering. *Journal of Fluency Disorders, 31*(4), 257–270. https://doi.org/10.1016/j.jfludis.2006.07.001

Howell, P., Rosen, S., Hannigan, G., & Rustin, L. (2000). Auditory backward-masking performance by children who stutter and its relation to dysfluency rate. *Perceptual and Motor Skills, 90*(2), 355–363. https://doi.org/10.2466/pms.2000.90.2.355

Hutchinson, J. M., & Ringel, R. L. (1975). The effect of oral sensory deprivation on stuttering behavior. *Journal of Communication Disorders, 8*(3), 249–258.

IBM Corp. (2015). *IBM SPSS statistics for Windows, Version 23.0.* Armonk, NY: Author.

Jones, R., Choi, D., Conture, E., & Walden, T. (2014). Temperament, emotion and childhood stuttering. *Seminars in Speech and Language, 35*(2), 114–131. https://doi.org/10.1055/s-0034-1371755

Karchmer, M., Mitchell, R. E., Marschark, M., & Spencer, P. E. (2003). Demographic and achievement characteristics of deaf and hard-of-hearing students. In M. Marschark & P. E. Spencer (Eds.), *Oxford handbook of deaf studies, language, and education* (pp. 21–37). New York, NY: Oxford University Press.

Kraft, S. J., & Yairi, E. (2012). Genetic bases of stuttering: The state of the art, 2011. *Folia Phoniatrica and Logopaedica, 64,* 34–47.

Lonigan, C. J., Wagner, R. K., Torgesen, J. K., & Rashotte, C. A. (2007). *Test of Preschool Early Literacy*. Austin, TX: Pro-Ed.

Månsson, H. (2000). Childhood stuttering: Incidence and development. *Journal of Fluency Disorders, 25,* 47–57.

Månsson, H. (2005). Stammens kompleksitet og diversitet [The complexity and diversity of the tribe]. *Dansk Audiologopædi, 41,* 13–33.

Max, L., Guenther, F., Gracco, V., Ghosh, S., & Wallace, M. (2004). Unstable or insufficiently activated internal models and feedback-biased motor control as sources of dysfluency: A theoretical model of stuttering. *Contemporary Issues in Communication Science and Disorders, 31,* 105–122.

McCreery, R. W., Walker, E. A., Spratford, M., Bentler, R., Holte, L., Roush, P., . . . Moeller, M. P. (2015). Longitudinal predictors of aided speech audibility. *Ear and Hearing, 36,* 24S–37S. https://doi.org/10.1097/AUD.0000000000000211

Moeller, M. P., & Tomblin, J. B. (2015). An introduction to the Outcomes of Children with Hearing Loss study. *Ear and Hearing, 36,* 92S–98S. https://doi.org/10.1097/AUD.0000000000000210

Moeller, M. P., Tomblin, J. B., & OCHL Collaboration. (2015). Epilogue: Conclusions and implications for research and practice. *Ear and Hearing, 36,* 4S–13S. https://doi.org/10.1097/AUD.0000000000000214

Montgomery, B., & Fitch, J. (1988). The prevalence of stuttering in the hearing-impaired school age populations. *Journal of Speech and Hearing Disorders, 53,* 131–135.

Ntourou, K., Conture, E. G., & Lipsey, M. W. (2011). Language abilities of children who stutter: A meta-analytical review. *American Journal of Speech-Language Pathology, 20*(3), 163–179. https://doi.org/10.1044/1058-0360(2011/09-0102)

Paden, E. P., Ambrose, N. G., & Yairi, E. (2002). Phonological progress during the first 2 years of stuttering. *Journal of Speech Language and Hearing Research, 45*(2), 256. https://doi.org/10.1044/1092-4388(2002/020)

Paden, E. P., Yairi, E., & Ambrose, N. G. (1999). Early childhood stuttering II: Phonology and stuttering. *Journal of Speech, Language, and Hearing Research, 42,* 1113–1124.

Proctor, A., Yairi, E., Duff, M. C., & Zhang, J. (2008). Prevalence of stuttering in African American preschoolers. *Journal of Speech Language and Hearing Research, 51*(6), 1465–1479. https://doi.org/10.1044/1092-4388(2008/07-0057)

R Core Team. (2016). *R: A language and environment for statistical computing*. Vienna, Austria: R Foundation for Statistical Computing. Retrieved from https://www.R-project.org/

Reilly, S., Onslow, M., Packman, A., Wake, M., Bavin, E., Prior, M., . . . Ukoumunne, O. C. (2009). Predicting stuttering onset by age of 3: A prospective, community cohort study. *Pediatrics, 123,* 270–277.

Sawilowsky, S. (2009). New effect size rules of thumb. *Journal of Modern Applied Statistical Methods, 8*(2), 467–474.

Smith, A., Goffman, L., Sasisekaran, J., & Weber-Fox, C. (2012). Language and motor abilities of preschool children who stutter: Evidence from behavioral and kinematic indices of nonword repetition performance. *Journal of Fluency Disorders, 37*(4), 344–358. https://doi.org/10.1016/j.jfludis.2012.06.001

Smith, A., & Weber, C. (2016). Childhood stuttering: Where are we and where are we going? *Seminars in Speech and Language, 37*(4), 291–297. https://doi.org/10.1055/s-0036-1587703

Sparrow, S. S., Cicchetti, V. D., & Balla, A. D. (2005). *Vineland Adaptive Behavior Scales–Second Edition*. Circle Pines, MN: American Guidance Service.

Spencer, C., & Weber-Fox, C. (2014). Preschool speech articulation and nonword repetition abilities may help predict eventual recovery or persistence of stuttering. *Journal of Fluency Disorders, 41,* 32–46. https://doi.org/10.1016/j.jfludis.2014.06.001

Stika, C. J., Eisenberg, L. S., Johnson, K. C., Henning, S. C., Colson, B. G., Ganguly, D. H., & DesJardin, J. L. (2015). Developmental outcomes of early-identified children who are hard of hearing at 12 to 18 months of age. *Early Human Development, 91*(1), 47–55.

Stiles, D. J., Bentler, R. A., & McGregor, K. K. (2012). The speech intelligibility index and the pure-tone average as predictors of lexical ability in children fit with hearing aids. *Journal of Speech, Language, and Hearing Research, 55*(3), 764–778. https://doi.org/10.1044/1092-4388(2011/10-0264)

Tomblin, J. B., Harrison, M., Ambrose, S. E., Oleson, J., & Moeller, M. P. (2015). Language outcomes in young children with mild to severe hearing loss. *Ear and Hearing, 36,* 76S–91S. https://doi.org/10.1097/AUD.0000000000000219

Tomblin, J. B., Walker, E. A., McCreery, R. W., Arenas, R. M., Harrison, M., & Moeller, M. P. (2015). Outcomes of children with hearing loss: Data collection and methods. *Ear and Hearing, 36,* 14S–23S. https://doi.org/10.1097/AUD.0000000000000212

Tourville, J. A., Reilly, K. J., & Guenther, F. H. (2008). Neural mechanisms underlying auditory feedback control of speech. *NeuroImage, 39*(3), 1429–1443. https://doi.org/10.1016/j.neuroimage.2007.09.054

van Lieshout, P., Peters, H., Starkweather, C., & Hulstijn, W. (1993). Physiological differences between stutterers and nonstutterers in perceptually fluent speech: EMG amplitude and duration. *Journal of Speech and Hearing Research, 36,* 55–63.

Weber-Fox, C., Wray, A. H., & Arnold, H. (2013). Early childhood stuttering and electrophysiological indices of language processing. *Journal of Fluency Disorders, 38*(2), 206–221. https://doi.org/10.1016/j.jfludis.2013.01.001

Wechsler, D. (2002). *Wechsler Preschool and Primary Scale of Intelligence–Third Edition*. San Diego, CA: Harcourt Assessment.

Wolk, L., Edwards, M. L., & Conture, E. G. (1993). Coexistence of stuttering and disordered phonology in young children. *Journal of Speech and Hearing Research, 36,* 906–917.

Yairi, E. (2013). Defining stuttering for research purposes. *Journal of Fluency Disorders, 38*(3), 294–298. https://doi.org/10.1016/j.jfludis.2013.05.001

Yairi, E., & Ambrose, N. (1992). A longitudinal study of stuttering in children: A preliminary report. *Journal of Speech and Hearing Research, 35,* 755–760.

Yairi, E., & Ambrose, N. (1999). Early childhood stuttering I: Persistency and recovery rates. *Journal of Speech, Language, and Hearing Research, 42,* 1097–1112.

Yairi, E., & Ambrose, N. (2005). *Early childhood stuttering*. Austin, TX: Pro-Ed.

Yairi, E., & Ambrose, N. (2013). Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders, 38,* 66–87.

Appendix

Fluency Survey

We would like to ask you some questions about your child's speech. These questions pertain to your child's speech right now, but also in the past, especially in the preschool years. We are interested in whether your child ever showed signs of stuttering. Below are examples of what stuttering is and what stuttering is not.

Stuttering is a disruption in the forward flow of speech that can take the form of repetitions, prolongations, or blocking. Below are examples of stuttered speech.

- Sound or syllable repetitions (e.g., *ba-ba-ba-ball* or *foot-foot-football*)
- Prolonged sounds (e.g., *s——ummer*)
- Silent blocks, typically at the beginning of words (e.g., ____*hat*)

Behaviors such as interjections (e.g., saying "um" or "like") or repeating whole words or phrases are not considered stuttering. Many children, especially during the preschool years, exhibit these types of behaviors, and they are not considered stuttering.

1) At any point in time did your child show signs of stuttering behavior (like those described above)?

(A) No, my child never displayed stuttering-like behaviors.

(B) Yes, my child stuttered for a period of time but he/she no longer does.

(C) Yes, my child is currently showing signs of stuttering.

If you answered A to question 1) you are done with this questionnaire. If you answered B or C to question 1) please continue.

2) Approximately what age did your child begin stuttering? __________

3) What types of stuttering behaviors did you observe (e.g., repetitions, prolongation, blocks)?

____

4) Did you notice any secondary behaviors (e.g., eye blinking, excessive tension) or avoidance behaviors (e.g., avoiding certain words, sounds, or speaking situations)?

(A) Secondary behaviors

(B) Avoidance behaviors

(C) Both secondary behaviors and avoidance behaviors

(D) Neither types of behaviors

Please elaborate if you answered A, B, or C. ___________________________________

5) Did your child ever receive treatment/services for stuttering?

A) Yes

B) No

If yes, please state what type of services and for how long. ______________________

If your child no longer stutters, at what age did the stuttering stop? __________

The Survey Presented in This Appendix Appears Courtesy of the Authors