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Research Article

Disfluency Characteristics of 4- and 5-Year-Old Children Who Stutter and Their Relationship to Stuttering Persistence and Recovery

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Purpose: The purpose of this study is to document disfluency behaviors expressed by 4- and 5-year-old children who stutter and to identify whether stuttering characteristics at this age are predictive of later stuttering recovery or persistence.
Method: We analyzed spontaneous speech samples from 47 children diagnosed with developmental stuttering when they were 4–5 years old. Based on their eventual diagnosis made the final year of participation in the longitudinal study when the children were 6–9 years old, the children were divided into two groups: children who eventually recovered from stuttering (n = 29) and children who were persisting (n = 18). We calculated a composite weighted stuttering-like disfluency (SLD) index of overall severity that considers the frequency, type, and number of repetition units of SLDs. The frequency and type of typical disfluencies were also examined.
Results: Higher weighted SLD scores at ages 4–5 years were associated with a higher probability of persistent stuttering. The weighted SLD also significantly discriminated between children who would eventually be diagnosed as persisting or recovered from stuttering. The frequency and type of typical disfluency did not distinguish the two groups of children; however, children who were persisting had significantly higher frequencies of part-word repetitions and dysrhythmic phonations (i.e., blocks, prolongations, and broken words) and maximum number of part-word repetitions compared to children who eventually recovered from stuttering.
Conclusions: Previous findings in younger, 2- to 3-year-old children who stutter did not suggest a relationship between the severity and type of children's SLDs and their eventual stuttering outcome. Yet, by the age of 4–5 years, we found that the weighted SLD, a clinically applicable tool, may be used to help identify children at greater risk for stuttering persistence. We propose that the weighted SLD be considered, along with other predictive factors, when assessing risk of stuttering persistence in 4- and 5-year-old children who are stuttering.

A substantial number (5%–11%) of children go through a period of stuttering in their preschool years (Andrews & Harris, 1964; Reilly et al., 2013; Yairi & Ambrose, 2005). Childhood onset fluency disorder, also referred to as "childhood stuttering," typically emerges in children around their third birthday, coinciding with marked advancement in children's speech and language abilities (Smith & Weber, 2017). Stuttering recovery rates are high (75%–80%) within the first 6–15 months of onset (Yairi & Ambrose, 1999, 2005); however, recovery rates decline to 50%–60% by the time a child reaches 5 years of age (Walsh et al., 2018; Yairi & Ambrose, 2005).

Given the number of preschoolers who begin to stutter, a significant concern is how to diagnostically differentiate children likely to recover from those at risk for developing chronic stuttering. In this prospective study, we examine disfluency characteristics of forty-seven 4- and 5-year-old children who stutter (CWS) divided into two cohorts based on their later stuttering outcomes: children eventually diagnosed as persisting (CWS-ePer) or recovered (CWS-eRec). Our goal is to determine if stuttering disfluency profiles are associated with a greater risk for eventual persistence. This is critical information to obtain as there are currently few

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predictive factors clinicians can rely upon to evaluate preschoolers' risk for stuttering persistence. One of these factors is family history—if CWS have a relative who persisted in stuttering, they are also more likely to persist (Walsh et al., 2018; Yairi & Ambrose, 2005). Another predictor is sex. Boys who stutter are more likely than girls to persist and develop a chronic stuttering disorder. The male-to-female ratio is approximately 2:1 near stuttering onset yet is estimated to be 5:1 by adulthood (Bloodstein & Ratner, 2008). Another predictor is age of stuttering onset. Preliminary data revealed that CWS who persisted, on average, began to stutter after 36 months, while CWS who recovered began to stutter slightly earlier, at 32.6 months; however, this trend was not statistically significant. (Yairi & Ambrose, 2005). Finally, the longer a child continues to stutter past 12 months onset, the lower the probability of recovery (Yairi & Ambrose, 2005). These factors serve as a guideline; however, they do not decisively identify individual children at risk for persistence.

The low accuracy in predicting stuttering persistence presents a significant barrier to prioritizing resources for those children who may need immediate therapy for stuttering. Delaying therapy for preschoolers at risk for persistence may permit maladaptive speech behaviors to form and places them at risk for developing negative feelings and attitudes toward communication (Clark et al., 2012; Vanryckeghem et al., 2005; Walsh et al., 2015; Yairi & Ambrose, 2005). It is vital then to discover additional clinically ascertainable factors associated with stuttering persistence to help determine a child's risk for developing chronic stuttering. Early intervention is critical, as therapeutic approaches have been shown to reduce stuttering-like disfluencies (SLDs), such as part-word (PW) repetitions, single-syllable (SS) whole-word repetitions, blocks, and prolongations, and help children maintain reduced levels of SLD (e.g., Millard et al., 2008; Onslow et al., 2003; Sonneville-Koedoot et al., 2015; Yaruss et al., 2006), as well as to provide emotional and educational support to the child and family (Yairi & Ambrose, 2005; Yaruss et al., 2006).

Diagnosing Stuttering in Preschool Children

SLD characteristics are used to diagnose stuttering in young children given these are a core feature of the disorder. In their efforts to establish guidelines to differentiate preschoolers who do and do not stutter, Ambrose and Yairi (1999) compared disfluency profiles from ninety 2- to 5-year-old CWS within 6 months of stuttering onset and 54 age-matched controls (children who do not stutter [CWNS]) and found that the frequency of SLD distinguished the two groups. CWS manifested significantly higher numbers of SLD compared to CWNS. On the other hand, speech sample analysis of typical disfluencies (TDs), such as multisyllabic word or phrase repetitions, filled pauses, or revisions, showed that the frequencies of TD did not differ between CWS and CWNS (Ambrose & Yairi, 1999; Yairi & Lewis, 1984; cf. Tumanova et al., 2014).

An additional characteristic that distinguishes CWS and CWNS is the extent of instances of stuttering, for example, the number of repetition units (RUs) or the number of times a segment of a word or a whole SS word is repeated within a single instance of stuttering (Ambrose & Yairi, 1995; Yairi & Lewis, 1984). Ambrose and Yairi (1995) reported that 33% of CWS repeated units 2 times or more (e.g., *b-b-but*) compared to only 13% of CWNS (Ambrose & Yairi, 1995). Moreover, Yairi (1983) found that parents of preschool CWS consider RUs to be among the most salient indicators that their child may be stuttering. Taken together, prior research confirms that stuttering behaviors in young CWS are reliably different from disfluent behaviors expressed by CWNS, leading to improved differential diagnosis of early childhood stuttering.

Disfluency Characteristics in Children Who Persist or Recover From Stuttering

Yairi et al. (1993) provided an early glimpse of disfluency characteristics of 2- and 3-year-old CWS within the first 12 weeks of onset. Their findings illustrated that SLD frequency near stuttering onset may be high, often peaking in severity at 2–3 months postonset, with SLD declining within 6 months postonset. Interestingly, frequencies of TD remained stable and did not show this trend. Later, by ages 3–4 years, Pellowski and Conture (2002) found an overall increase in SLD, with the frequency of dysrhythmic phonations (DPs), in particular, showing a significant increase among 4-year-old CWS compared to 3-year-old CWS. These fluctuations in stuttering severity near onset are unsurprising given the ongoing development of the neural systems supporting speech and language functions and that some preschoolers in the group of CWS may already be in the process of recovery. Necessarily, children who will eventually recover will experience a decline in frequency of SLD to the level where they no longer qualify as stuttering. Nevertheless, there have been few prospective studies comparing early disfluency characteristics from children for whom the eventual persistence/recovery was documented.

In a longitudinal study of stuttering behaviors, Yairi and Ambrose (1992) followed twenty-seven 2- to 4-year-old children for up to 2 years collecting speech samples at regular intervals. They noted an overall decline in SLD and TD over time in all CWS, but children who would eventually develop persistent stuttering (CWS-ePer; n = 9) had significantly higher stuttering rates (SLD/100 syllables) than children who would eventually recover (CWS-eRec; n = 18). Differences between groups of children who persisted or recovered became evident by approximately 20 months postonset. Follow-up studies from this research group (Throneburg & Yairi, 2001; Yairi et al., 1996) did not replicate this result. Thus, this group did not draw firm conclusions from their data about the usefulness of disfluency analysis for 3- and 4-year-old CWS for predicting the child's ultimate stuttering status.

To summarize, research reveals that the frequency of SLD in child-onset stuttering is variable near the onset of the disorder, but a general decline in SLD over time occurs for many CWS (Yairi & Ambrose, 1992; Yairi et al., 1993). Longitudinal studies suggest that near the onset of stuttering, the severity and type of SLD can be similar between CWS-eRec and CWS-ePer, but between-groups differences clearly must emerge over time, as these children mature toward eventual persistence or recovery. The few longitudinal studies of stuttering persistence and recovery from Yairi and colleagues have included modest sample sizes and provided important preliminary data and descriptive statistics (Yairi & Ambrose, 1992; Yairi et al., 1996). To date, no published studies have examined the relationship between SLD characteristics and stuttering recovery or persistence for older cohorts (4 and 5 years old) of CWS. The 4- to 5-year period is a pivotal stage in terms of eventual recovery and persistence, because the average age at onset of stuttering is around 3 years, and the majority of recovery occurs within 15 months postonset (Yairi & Ambrose, 1999, 2005). It follows that, on average, many CWS are referred to a speech-language pathologist (SLP) for concerns about stuttering at approximately 55 months (Yaruss et al., 1998). Clinicians evaluating preschool CWS must make consequential decisions regarding treatment within this developmental window (Yairi et al., 1996), as awareness of stuttering is emerging along with the potential for adverse impact (Boey et al., 2009; Langevin et al., 2010; Vanryckeghem et al., 2005). The goal of this study is to document disfluency behaviors expressed by 4- and 5-year-old CWS and to identify whether stuttering characteristics at ages 4 and 5 years are predictive of these children's eventual stuttering recovery and persistence determined when they are 6–9 years old.

We use a comprehensive clinical index initially developed to diagnose stuttering in young children, the weighted SLD (Ambrose & Yairi, 1999). The weighted SLD calculates three dysfluency dimensions: the type and frequency of SLD (PW repetitions, SS whole-word repetitions, and dysrhythmic productions—DP blocks, broken words, and prolongations) and the average number of RUs into one score. We investigate the weighted SLD's potential for predicting the probability for eventual persistence of stuttering in preschool CWS to assess the hypothesis that stuttering behaviors at age 4–5 years may be used to help distinguish children eventually diagnosed as persisting from children eventually diagnosed as recovered.

Method

Participants

This study utilized data gathered as part of a larger, dual-site, longitudinal study examining risk factors for persistent stuttering in preschool children. In this study, we focus on analyses of the spontaneous speech samples from children for whom an eventual stuttering status (i.e., either persistence or recovery) was obtained.

Data were collected at Purdue University and The University of Iowa using identical testing and language sampling protocols. The research protocols were conducted with the approval of the institutional review boards of both universities. Speech samples were analyzed from a group of 47 preschool CWS (11 girls and 36 boys) for whom an eventual diagnosis of stuttering recovery or persistence status could be determined when the child was older (school-age). We also analyzed speech samples from a group of twenty-seven 4- and 5-year-old CWNS (eight girls and 19 boys) to provide a normative reference of the disfluency characteristics expressed by typically speaking preschool children. Data from the group of CWNS, however, were not included in the statistical analysis. All CWS were between the ages of 4 and 5 years, ranging from 48 to 71 months. The 47 participants comprised two groups: 29 children who would eventually be diagnosed as recovered from stuttering (CWS-eRec) and 18 children who would eventually be diagnosed as persisting (CWS-ePer; see the Appendix). All participants spoke American English as their primary language, passed a standard hearing screening (500, 1000, 2000, 4000, and 6000 Hz at 20 dB HL), had normal or corrected-to-normal vision, no history of neurological problems or serious illnesses, and were not receiving medication expected to affect the central nervous system. All participants in the study had nonverbal/reasoning skills within normal limits as assessed by the Columbia Mental Maturity Scale (Burgemeister et al., 1972).

Determination of stuttering status was consistent with Yairi and Ambrose's (1999) diagnostic criteria. Participants included in the CWS group met these four criteria at the onset of the study: (a) the parent(s) considered the child to be stuttering; (b) the project SLP (fourth author) considered the child to be stuttering; (c) the child received a stuttering severity rating of 2 or higher on a 0–7 severity scale developed by Yairi and Ambrose, with 0–1 indicating *normal fluency/borderline*, 2–3 indicating *mild stuttering*, 4–5 indicating *moderate stuttering*, and 6–7 indicating *severe stuttering* from either a parent or the SLP; and (d) the child exhibited three or more SLDs per 100 syllables collected across two conversational speech samples. See the Appendix for each participant's syllable count. In 10 cases (seven CWS-eRec, three CWS-ePer),[1] the child's SLD did not reach the minimum 3% SLD criteria, yet the parent considered their child to be stuttering and reported the speech sample was inconsistent with their child's typical severity at home and in other contexts. In these cases, the project SLP assessed the child's fluency during other interactions throughout the laboratory visit, for instance, during

[1]The TD and SLD measures based on groupings that included only those CWS who had at least three SLDs per 100 syllables in their spontaneous language sample at their initial visit were also analyzed. We found no difference in the outcome for any statistical test or model between these results and the results reported here for the larger *N*, more inclusive groupings. Therefore, for succinctness, the findings for the smaller subset of CWS (15 CWS-ePer, 22 CWS-eRec) are not presented in detail.

greetings/departures, laboratory tours, activity transitions, and other assessments. The SLP made clinical judgments regarding the presence and frequency of stuttering following observations of increased SLD in these additional contexts. This is reasonable given the variability of stuttering in different situations (Constantino et al., 2016; Yaruss, 1997). It is critical to include a sample that represents the population clinicians see and assess, often over a single diagnostic session.

Participants returned to the laboratory each year to have their speech reevaluated in this longitudinal study. A child was classified as recovered if they met the following criteria: (a) the parent(s) no longer considered the child to be stuttering, (b) the project SLP no longer considered the child to be stuttering, (c) the child received a stuttering severity rating of less than 2 from both a parent and the project SLP, and (d) the child exhibited less than three SLDs per 100 syllables collected during the two spontaneous speech samples. If a child who stutters did not meet all these criteria, then they were considered persisting. We used a child's status from their final year of participation for persist/recover classification for this study. The Appendix provides the age and yearly visit for each child's final visit.

Speech Sample Collection

The speech samples used in our analysis were obtained when children were between 4 and 5 years and diagnosed as stuttering. We audio- and video-recorded children's spontaneous speech during two play-based sessions. In the first session, the child played with a familiar caretaker, typically a parent with a Playmobil set. The project SLP encouraged parents to pause often to allow their child to speak and to ask open-ended questions to elicit longer productions. The second speech sample was recorded while the child and the SLP engaged in conversation while playing with Play-Doh. The SLP had obtained a list of topics suggested by the parent that would be of interest to the child and stimulate more productions. Topics typically covered relevant events, such as birthdays, holidays, movies, and vacations.

Speech Sample Transcription

Samples were orthographically transcribed and coded using the Systematic Analysis of Language Transcripts program (Miller & Iglesias, 2006). Each sample was analyzed twice. During the first analysis, the trained graduate clinician transcribed each utterance and coded for all disfluencies. During training, graduate clinicians had to achieve at least 90% reliability with the project SLP on practice transcripts identifying the presence of disfluencies and achieve at least 80% reliability coding the specific type of SLDs and TDs. These included PW repetitions, SS whole-word repetitions, DPs including blocks, broken words, and prolongations. The number of PW and SS repetitions was also coded (e.g., *b-baby* would be coded as [PW], while *i-i-it* would be coded as [PW2]). Other (typical) disfluencies were designated with the code "TD," including interjections (I), revisions (R), phrase repetitions (P), and multisyllabic word repetitions (M). The project SLP then analyzed the samples a second time. Any disagreements about the type of disfluency or the number of repetitions in the coded speech samples were resolved by the SLP and the graduate student listening together to the segment in question to come to a consensus.

Following the final analysis, a syllable count was performed for each transcription set in order to normalize disfluency measures. To standardize the syllable range, a maximum of 1,210 syllables was included. If a participant's sample exceeded 1,200 syllables, the transcript was truncated at the end of the utterance in which the 1,200-syllable limit was achieved, resulting in a maximum of 1,210 syllables. We attempted to obtain a comparable number of syllables from each speech sample (e.g., 600 syllables from the child–parent sample and 600 syllables from the child–examiner sample), although this could not be accomplished for all children. Only one child's sample failed to reach the 600-syllable limit (only 264 were included) due to severity of stuttering (see the Appendix).

Although all utterances were transcribed, specific elements were excluded from the speech sample analysis in order to avoid skewing the syllable counts. The excluded elements included rote language performances (such as counting or reciting nursery rhymes), unintelligible utterances, direct repetitions of adult utterances, revisions or phrase repetitions, SLDs or TDs, and single-word responses to questions (e.g., yes, no, okay).

Data Analysis

We analyzed disfluency data for each child in the study who provided a speech sample at either 4 or 5 years of age. For children who provided speech samples at ages 4 and 5 years, we analyzed the earlier sample to maintain the closest proximity to the onset of stuttering. Both SLD and TD codes were then tallied and entered into a database in order to calculate our variables. Disfluency types were computed as the percentage of the total number of syllables from each participant's speech sample set. To explore the potential link between eventual stuttering status and repetitions, we analyzed the average number of PW and SS repetitions (this value was used in the weighted SLD calculation described below), along with the maximum number of PW and SS repetitions from each speech sample. Finally, we calculated a comprehensive measure of stuttering severity, the weighted SLD, for each participant (Ambrose & Yairi, 1999). The weighted SLD is computed by summing PW and SS repetitions per 100 syllables of speech and then multiplying this value by the mean number of PW and SS RUs combined. This value is added to twice the sum of blocks and prolongations (collectively called as DPs). Because DP rarely occur in typical speakers and infrequently occur in early childhood stuttering (Yairi & Ambrose, 2005), they receive a weighting (i.e., multiplied by 2). The resulting equation is: [(PW + SS) × mean RU) + (2 × DP) = weighted SLD]. A score of 4.0 or higher is used to distinguish CWS from typically fluent children. Note that the weighted SLD may be computed using FLUCALC, a profiling command available in CHILDES CLAN software (MacWhinney, 2000).

Statistical Analysis

In accordance with study criteria, the disfluency profiles of CWNS would necessarily be different from the groups of children diagnosed as stuttering; therefore, no statistical analysis of stuttering characteristics included the CWNS group. However, we include data from CWNS in Table 1 as a reference. The focus of the current study is on 4- to 5-year-old CWS who are eventually diagnosed as persisting or recovered. We conducted independent-samples *t* tests ($p < .05$) to compare age in months at study onset, age at stuttering onset, duration of stuttering at onset, and age at final study visit for the CWS-eRec and CWS-ePer groups.

Disfluency frequency data are usually not normally distributed as they often comprise percentages and counts within a narrow range (e.g., between 1% and 10%) and may include zero values (Jones et al., 2006; Tumanova et al., 2014). Visual inspection of distributions and Shapiro–Wilk tests of normality confirmed that disfluency variables were right-skewed. We thus calculated nonparametric Mann–Whitney *U* tests to assess potential differences between CWS-eRec and CWS-ePer in the frequency of subcategories of SLD (PW repetitions, SS word repetitions, and DPs), frequency of subcategories of TD (interjections [I], revisions [R], multisyllabic word repetitions [M], and phrase repetitions [P]), the average number of repetitions for PW and SS combined, and the maximum number of PW and SS repetitions. Bonferroni-corrected alpha level was set at $p \leq .017$ for the three categories of SLD, $p \leq .012$ for the four categories of TD, and $p \leq .017$ for the three repetition calculations. Finally, effect sizes were calculated for each Mann–Whitney *U* test using the formula, $r = \frac{Z}{\sqrt{n}}$. Cohen's $r = .1$ represents a small effect, $r = .3$ represents a medium effect, and $r = .5$ represents a large effect size.

The weighted SLD is derived from the other dependent variables comprising the type, frequency, and number of repetitions of SLD. We examined the relationship between the weighted SLD and stuttering persistence and recovery with two analyses. First, we conducted a binary logistic regression model to examine whether there was a statistically significant relationship between the weighted SLD and stuttering outcome (i.e., persistence or recovery). This was computed using R Studio (RStudio Team, 2019), a companion program to R (R Core Team, 2019). R packages were used for data management, analysis, and visualization (car: Fox & Weisberg, 2019; ggplot2: Wickham, 2016; tidyr: Wickham & Henry, 2019). Odds ratios were calculated from logits and then transformed into probabilities for ease of visualization and interpretation. This continuous prediction (weighted SLD) of the binary outcome (i.e., persisting or recovering) was then assessed.

Next, we calculated a receiver operating characteristic (ROC) curve analysis that revealed the diagnostic ability of the weighted SLD to classify persistence and recovery by providing the trade-off between the weighted SLD's sensitivity versus 1 − specificity. In ROC analysis, the true positive (TP) rate is plotted against the false positive (FP) rate. The TP rate is the proportion of CWS-ePer correctly predicted to be persisting (i.e., TPs) out of all persisting children (TP / TP + false negatives), while the FP rate is the proportion of CWS-eRec who were incorrectly predicted to be persisting or FPs out of all recovered children (FP / true negative + FP). The area under the ROC curve was used to assess the ability of the weighted SLD to classify stuttering persistence and recovery. An area under the ROC curve of 0.5 suggests no discriminative ability (e.g., chance/coin toss), while 0.7–0.8 are considered acceptable, 0.8–0.9 are considered excellent, and 0.9 and above are considered outstanding discriminative ability (Mandrekar, 2010).

Table 1. Means, standard deviations (*SD*), and 95% confidence intervals (CI) for percent part-word repetitions (%PW), percent single-syllable word repetitions (%SS), percent dysrhythmic phonations (%DP), average PW and SS repetition units (Avr RUs), maximum PW and maximum SS repetition units (RUs), weighted stuttering-like disfluency (SLD) index, and typical disfluencies (%TD).

	SLDs							
Group	**%PW (*SD*) [95% CI]**	**%SS (*SD*) [95% CI]**	**%DP (*SD*) [95% CI]**	**Avr RU (*SD*) [95% CI]**	**Max PW RU (*SD*) [95% CI]**	**Max SS RU (*SD*) [95% CI]**	**Weighted SLD (*SD*) [95% CI]**	**%TD (*SD*) [95% CI]**
CWS-ePer	2.88 (2.22) [1.78, 3.99]	2.69 (2.01) [1.69, 3.69]	2.41 (2.97) [.93, 3.89]	1.44 (.35) [1.27, 1.62]	4.28 (2.32) [3.12, 5.43]	3.89 (2.76) [2.51, 5.26]	13.36 (8.54) [9.11, 17.61]	3.93 (2.57) [2.66, 5.21]
CWS-eRec	1.47 (1.09) [1.06, 1.37]	1.95 (1.15) [1.51, 2.39]	.89(.76) [.60, 1.18]	1.25 (.32) [1.13, 1.37]	2.83 (1.69) [2.18, 3.47]	3.07 (1.51) [2.49, 3.64]	6.28 (4.19) [4.69, 7.88]	3.95 (1.84) [3.25, 4.65]
CWNS	.32 (0.25) [.22, .42]	.55 (0.46) [.36, .73]	.20 (.21) [.12, .29]	1.29 (1.0) [.88, 1.70]	1.15 (.66) [.89, 1.41]	1.30 (.67) [1.03, 1.56]	1.33 (0.82) [1.01, 1.65]	3.21 (1.18) [2.74, 3.68]

Note. CWS-ePer = children who eventually persisted in stuttering; CWS-eRec = children who eventually recovered from stuttering; CWNS = children who do not stutter.

Results

Demographics

There was not a significant difference in age (in months) between CWS-ePer (M = 58.4, SD = 7.8) and CWS-eRec (M = 54.7, SD = 5.3) at the initial visit (when all children were between ages 4 and 5 years and diagnosed as stuttering), $t(45)$ = 1.92, p = .06. Importantly, CWS-ePer (M = 36.3, SD = 9.8) and CWS-eRec (M = 34.3, SD = 8.1) had a statistically similar reported age at stuttering onset, $t(45)$ = 0.76, p = .45, and had been stuttering for a comparable length of time (CWS-ePer: M = 22.1, SD = 8.7; CWS-eRec: M =20.4, SD = 58.5), $t(45)$ = 0.65, p = .52. Finally, there was no significant difference in age between CWS-ePer (M = 95.3, SD = 10.1) and CWS-eRec (M = 91.7, SD = 11.2) at the final visit of the study (when final status was determined), $t(45)$ = 1.10, p = .28. The Appendix provides each participant's age at Visit 1, age at stuttering onset, duration of stuttering at Visit 1, and their age and last year of participation in the longitudinal study.

SLDs

Weighted SLD Index

The means and standard deviations of the weighted SLD for each group are summarized in Table 1. Results from the bivariate logistic regression indicated that the weighted SLD significantly predicted persistence in stuttering (B = .18, p = .004, OR = 1.20, 95% CI [1.08, 1.38]). This indicates that, for each 1-unit increase in weighted SLD score, a child's odds of persisting in stuttering also increased by a factor of 1.20. The predicted probability of persistence is visualized in Figure 1.

The calculation of sensitivity and specificity of selected cutoff values of the weighted SLD for predicting stuttering persistence and recovery are shown in Table 2, along with the number of CWS-ePer and CWS-eRec correctly and incorrectly diagnosed at each cutoff value. The corresponding ROC curve, which represents the weighted SLD index (blue line), is shown in Figure 2 and based on a nonparametric method using IBM SPSS Statistics, Version 26. The closer the ROC curve is to the top left corner, the more efficient the test is, while ROC curves that fall on the diagonal line (area under ROC of 0.5 or chance) offer no predictive value. The area under the ROC curve for predicting stuttering persistence was 0.77 (SE = 0.07, 95% CI [0.62, 0.91], p = .002). The ROC curve and corresponding area under the ROC curve value reveal that the weighted SLD had acceptable predictive ability to discriminate between CWS who were eventually diagnosed as persisting from children eventually diagnosed as recovered from stuttering.

SLD Type and Frequency

Table 3 provides Spearman rho correlations between the weighted SLD and its constituent components. As expected, we found moderate to strong (r_s = .62–.80) correlations between the weighted SLD and the individual SLD characteristics (PW repetitions, SS whole-word repetitions DPs, and average RUs). We also found moderate correlations (r_s = .42–.62) among the SLD characteristics. Mann–Whitney U tests indicated that the frequency of PW repetitions (U = 147, p = .013, r = −.36) and DP (U = 150, p = .015, r = −.35) were significantly higher (each representing a medium effect) for the CWS-ePer (Mdn PW = 2.36, Mdn DP = 1.34) compared to the CWS-eRec (Mdn PW = 1.33, Mdn DP = 0.75). However, the difference in the frequency SS repetitions between CWS-ePer (Mdn = 2.09) and CWS-eRec (Mdn = 1.76) was not significant (U = 209, p = .26, r = −.17). Hence, the percentage of PW repetitions and DPs differentiated groups of 4- and 5-year-old CWS who eventually persisted or recovered. The means, standard deviations, and confidence intervals of the difference of the frequencies of each type of SLD for each group are provided in Table 1.

SLD Repetitions

Table 1 also provides the mean number of PW and SS word RUs (i.e., the value used in the weighted SLD calculation) and the maximum number of repetitions (i.e., the maximum number of times a child repeated a stuttered syllable in a single occurrence) of PW and SS. We did not detect a group difference in average PW and SS repetitions (CWS-ePer Mdn = 1.34, CWS-eRec Mdn = 1.24; U = 178, p = .07, r = −.26). However, the maximum number of PW repetitions was significantly higher for CWS-ePer (Mdn = 4.0) compared to CWS-eRec (Mdn = 2.0; U = 151.5, p = .01, r = −.36). There was no significant group difference in maximum number of SS repetitions (CWS-ePer Mdn = 3.0, CWS-eRec Mdn = 3.0; U = 233, p = .53, r = −.09).

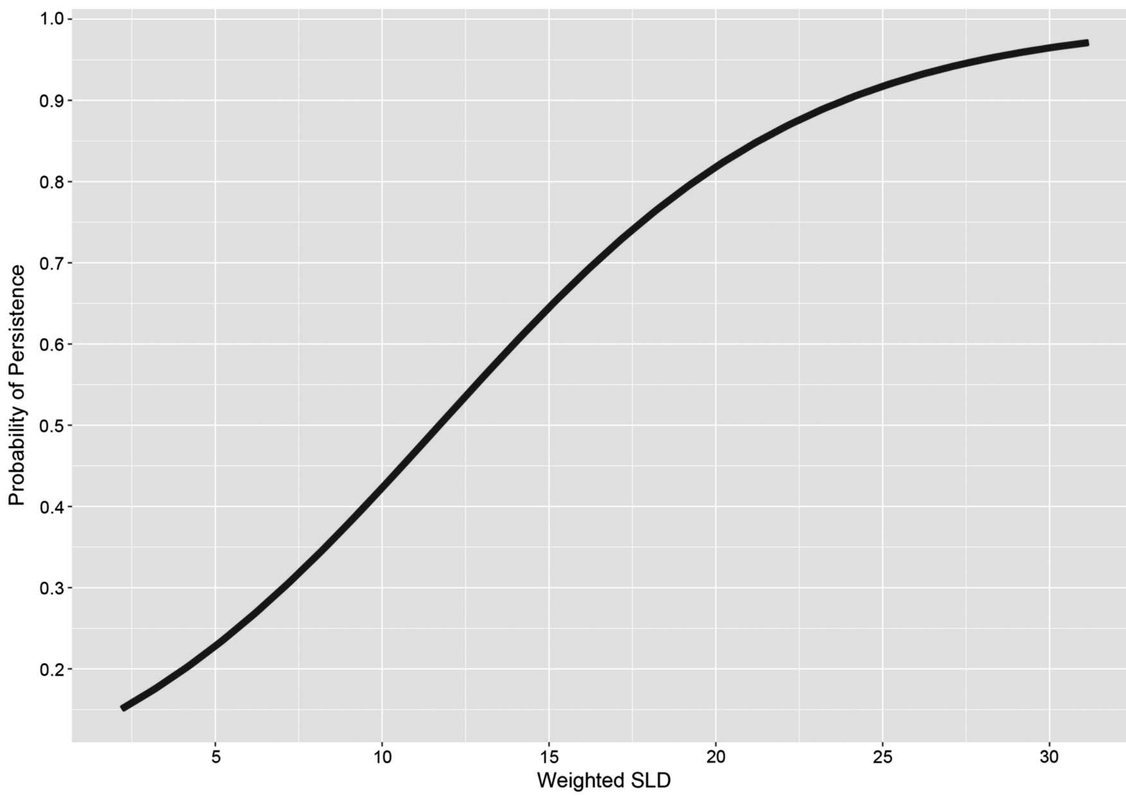
TDs

Mann–Whitney U tests indicated that the two groups had statistically similar frequencies of TD: interjections (CWS-ePer Mdn = 1.42, CWS-eRec Mdn = 1.48; U = 238, p = .62, r = −.07), revisions (CWS-ePer Mdn = 1.17, CWS-eRec Mdn = 1.32; U = 212.5, p = .28, r = −.16), multisyllabic word repetitions (CWS-ePer Mdn = .08, CWS-eRec Mdn = .08; U = 226, p = .43, r = −.11), and phrase repetitions (CWS-ePer Mdn = .63, CWS-eRec Mdn = .66; U = 235, p = .57, r = −.08). We collapsed across the subtypes of TD to report the means, standard deviations, and confidence intervals for each group (see Table 1).

Discussion

The primary aim of this study was to determine whether speech disfluency characteristics displayed by 4- and 5-year-old CWS are predictive of eventual stuttering persistence and recovery determined when children were of school age at 6–9 years. We calculated a weighted SLD index reflecting the type, frequency, and number of repetitions of SLD and assessed the ability of this composite

Figure 1. Weighted stuttering-like disfluency (SLD) scores as a function of the probability of stuttering persistence. A 1-unit increase in weighted SLD score was associated with a 1.20 increase in the probability of stuttering persistence in 4- and 5-year-old children who stutter.



measure to predict stuttering persistence and recovery. Higher weighted SLD scores at ages 4–5 years were associated with a higher probability of stuttering persistence. The ROC curve analysis suggested that the weighted SLD also had acceptable discriminative ability to differentiate between children who would eventually be diagnosed as persisting or recovered from stuttering.

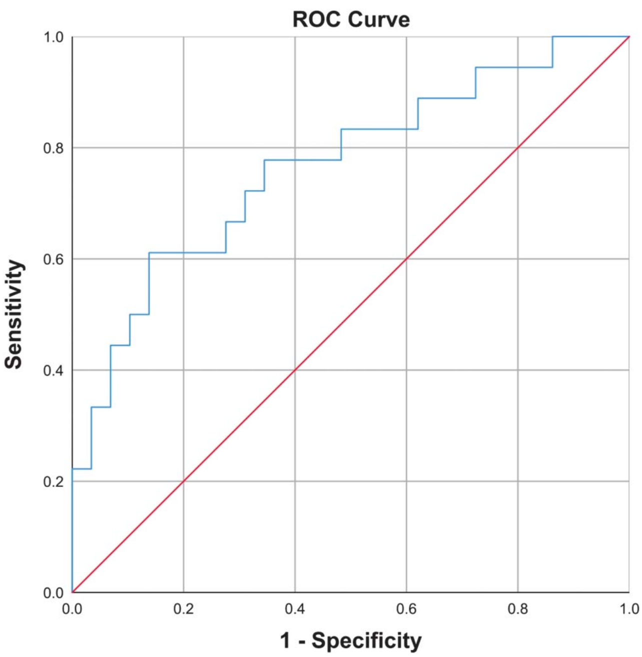
We also examined group differences in stuttering characteristics, including PW and SS whole-word repetitions, DPs, and the average and maximum number of

Table 2. Sensitivity and 1 – specificity expressed as percentages at different cutoff values of the weighted stuttering-like disfluency (SLD; calculated using speech samples from 4- and 5-year-old children who stutter when they were diagnosed as stuttering) for predicting eventual stuttering persistence.

Weighted SLD cutoff values	CWS-ePer (*n* = 18)		CWS-eRec (*n* = 29)			
	True positive	False negative	false Positive	True negative	Sensitivity	1 – specificity
2.53	18	0	25	4	100%	86.2%
3.58	16	2	21	8	88.9%	72.4%
4.19	15	3	18	11	83.3%	62.1%
5.26	15	3	14	15	83.3%	48.3%
6.26	14	4	10	19	77.8%	34.5%
6.83	13	5	9	20	72.2%	34.5%
7.94	11	7	8	21	61.1%	27.6%
9.66	11	7	4	25	61.1%	13.8%
10.85	9	9	4	25	55.6%	13.8%
12.93	8	10	3	26	44.4%	10.3%
16.75	6	12	1	28	33.3%	03.4%
19.53	4	14	0	29	22.2%	0%
29.10	1	17	0	29	05.6%	0%

Note. CWS-ePer = children who eventually persisted in stuttering; CWS-eRec = children who eventually recovered from stuttering.

Figure 2. Receiver operating characteristic (ROC) curve of the weighted stuttering-like disfluency (blue line) to discriminate eventual stuttering recovery and persistence. True positive rate or sensitivity is plotted along the *y*-axis against false positive rate or 1 – specificity on the *x*-axis. The 45° red diagonal line serves as a null reference denoting no discrimination.



repetitions. The frequency of PW and DP and mean maximum number of PW repetitions were significantly higher in CWS-ePer than in CWS-eRec, while the frequency of SS, average RU score for PW and SS combined, and maximum number of SS repetitions were statistically similar for the CWS-eRec and CWS-ePer groups.

Earlier findings suggest that CWS and CWNS produce similar frequencies of TDs (speech disfluencies made by people who do and do not stutter) in their speech (Ambrose & Yairi, 1999; Pellowski & Conture, 2002; Yairi & Ambrose, 2005; Yairi & Lewis, 1984). We found that the frequency of TD in speech samples collected from CWS at ages 4–5 years did not distinguish the CWS-ePer and CWS-eRec groups.

Table 3. Spearman rho correlations between weighted stuttering-like disfluency (SLD) and constituent variables (n = 47).

Variable	PW	SS	DP	Avr RU
Weighted SLD	.803**	.663**	.748**	.621**
PW		.422**	.624**	.510**
SS			.241	.524**
DP				.217

Note. PW = part-word repetitions; SS = single-syllable whole-word repetitions; DP = dysrhythmic phonations; Avr RU = average repetition unit

**p < .01 (two-tailed).

Finally, we did not find significant group differences in the demographic risk factors age at stuttering onset and duration of stuttering. Yairi and Ambrose (2005) reported that CWS-ePer began stuttering, on average, approximately 3.5 months later than CWS-eRec, although this difference was not statistically significant. Our cohort of CWS-ePer began stuttering approximately 2 months later than the group of CWS-eRec; however, there was considerable overlap between the two groups. Yairi and Ambrose also noted that the chance of recovery decreases after 1 year of stuttering. Most of our 4- to 5-year-old participants had already been stuttering well over 1 year when they entered the study (CWS-ePer M = 22.1 months, CWS-eRec M = 20.4 months). Thus, the overall stuttering recovery rate for our participants was lower, approximately 62%, compared to 75%–80% recovery rates reported within the first 6–15 months after onset (Yairi & Ambrose, 1999, 2005). Our sample does not include those children whose stuttering resolved early (i.e., between the ages of 2 and 3 years). Therefore, we suggest that the duration of stuttering in 4- and 5-year-old CWS may not be as useful as a risk indicator as it is for CWS at younger or older ages.

Weighted Stuttering Index

The weighted SLD is a composite measure developed to distinguish normal disfluency from stuttering in preschoolers (Ambrose & Yairi, 1999). It synthesizes the type, frequency, and number of repetitions of SLD into one score. A weighted SLD score of 4.00–9.99 indicates mild stuttering, a score of 10.00–29.99 indicates moderate stuttering, and a score of ≥ 30.00 indicates severe stuttering. In our cohort of 4- and 5-year-old CWS, 15 out of 18 or 83% of CWS-ePer scored above the median (4.96) of CWS-eRec, while only three out of 29 or 10% of CWS-eRec scored above the median (11.62) of CWS-ePer. Bivariate logistic regression revealed that a 1-unit increase in weighted SLD score was associated with a 1.20 increase in the odds of persisting in stuttering. Odds ratios were converted into probabilities for ease of interpretation in Figure 1. A child's score can be compared to this curve to assess the potential probability of persistence. For example, the curve reveals that a weighted SLD score at the cutoff for mild stuttering, or approximately 10.00, is associated with up to an approximately 43% chance of persistence, while the chance of persistence increases to 65% for a moderate severity score of 15.

We conducted an ROC curve analysis to assess the ability of the weighted SLD to distinguish between those children who would eventually be diagnosed as persisting or recovered from stuttering. In ROC curve analysis, the ideal cutoff score has the highest TP rate and the lowest FP rate. From a clinical perspective, however, it would not be prudent to recommend a single cutoff value for clinicians and parents to rely on when making consequential decisions regarding treatment for several reasons. In the case of early intervention, failing to identify a TP may have profound, lifelong ramifications. On the other hand, recommending treatment for a child who would have recovered without treatment, albeit costly and an expenditure of scarce resources, may be a more acceptable trade-off with less impactful consequences. Second, clinicians should always consider multiple factors when making therapy recommendations, a point we return to in the conclusion. For example, if a child is male and/or has a positive family history of stuttering, a clinician may wish to adopt a more conservative cutoff score. In this case, a weighted SLD of 5.26 would identify 83.3% of children at risk for persistence yet would also incorrectly identify a substantial number (48.3%) of children who would recover naturally. If a child had no risk factors, a weighted score of 6.83 would identify approximately 78% of children at risk for persistence and have a lower FP rate, incorrectly identifying 35% of children who would recover.

There are a few issues that are important to acknowledge. One is that our findings are based on 47 children, 18 who are persisting and 29 diagnosed as recovered from stuttering. Clearly, replicating and bolstering these results with longitudinal data from additional preschool CWS would further support the use of the weighted SLD to help assess a child's risk for persistence. Another issue to consider is that some participants' stuttering status may still be in a transitory phase. It is possible, for example, that some of the younger participants who are persisting may eventually recover. Despite these issues, we provide empirical support for the weighted SLD index, a clinically useful and accessible measure, to help identify risk for stuttering persistence in 4- and 5-year-old children.

SLD Characteristics

Our analysis of SLD characteristics in 4- and 5-year-old CWS revealed that the CWS who would eventually persist in stuttering (CWS-ePer) displayed higher frequency of PW and DP subcategories of SLD. How do these findings expand upon a body of evidence from younger children suggesting disfluency measures are not predictive of eventual status? A careful examination of the literature suggests that the answer may be a matter of developmental changes.

As discussed, previous research has provided us with a developmental picture of the frequencies of SLD for both CWS-eRec and CWS-ePer near the time of onset. Findings suggest that the majority of CWS, regardless of eventual status, initially experience a peak in the frequency of SLD between 2 and 3 years of age (Yairi & Lewis, 1984)—likely within the first several months of stuttering onset (Yairi et al., 1993). SLD frequency shows a general decline the following year after onset for many CWS (Yairi & Ambrose, 1992; Yairi et al., 1993). Pellowski and Conture (2002) suggested a potential increase in disfluencies between the ages of 3 and 4 years, though the design of their study did not allow the distinction of the CWS-ePer and CWS-eRec groups. The few preliminary studies that accounted for eventual status, however, documented that many CWS-eRec continued to experience a general decline following the peak postonset, while CWS-ePer, on average, did not manifest this rate of decline (Yairi & Ambrose, 1992; Yairi et al., 1996). However, the difference in SLD frequency between the CWS-ePer and CWS-eRec groups did not reach levels of significance in studies focusing primarily on younger children up to the age of 4 years (Yairi & Ambrose, 1992; Throneburg & Yairi, 2001). Our results reveal that, by 4–5 years, SLD characteristics of CWS-ePer and CWS-eRec have diverged significantly enough to be captured through clinical measures. Our findings, therefore, build upon, rather than contradict, previous findings, contributing to a clearer picture of the differences in SLD characteristics between CWS-ePer and CWS-eRec through the developmental window of the preschool years when most children are likely to experience both the onset and recovery from stuttering. However, we note that the frequency of SS whole-word repetitions did not distinguish the CWS-ePer and CWS-eRec groups, indicating that frequency of SS is not specifically associated with chronic stuttering at this age.

Ambrose and Yairi (1995) reported that RUs greater than 1.5 reliably differentiate children who do and do not stutter. The average number of PW and SS repetitions, which is the value used in the weighted SLD calculation, was similar between the CWS-ePer and CWS-eRec groups. Interestingly, the mean number of maximum PW repetitions for the CWS-ePer group (approximately 4) was significantly higher than the mean number for the CWS-eRec group (approximately 3). Analyzing maximum RUs allowed us to highlight disfluency characteristics on the more extreme end of the severity continuum. Therefore, a clinician cannot assume a child who will persist in stuttering will exhibit four or more repetitions during each stuttering event. However, the presence of PW repetitions of four in a 4- to 5-year-old child who stutters may serve as an additional risk indicator for persistent stuttering. The maximum number of repetitions for SS repetitions did not distinguish the CWS-eRec and CWS-ePer groups, again signifying that SS disfluencies may not reliably differentiate children who persist or recover from stuttering. A potentially valuable follow-up experiment might use these findings to guide a recalibration of the weighted SLD for predicting stuttering persistence in 4- to 5-year-olds. For example, the discriminative ability of this index might improve if the repetition component focused exclusively on PW or perhaps weighted PW more than SS.

An important point to highlight before closing the discussion on SLD characteristics is the significant moderate relationships among the different SLD characteristics and the significant moderate-to-strong relationships, as expected, among SLD characteristics and the weighted SLD (see Table 3). Thus, although we observed significant differences between the groups with respect to several SLD characteristics, care should be taken when interpreting the results of group comparisons of individual SLD characteristics given the interdependence among them.

Considerations and Conclusions

Established factors associated with stuttering persistence (e.g., age at onset, time since onset, family history, sex), along with the present findings regarding SLD characteristics, are revealed by comparing across groups of CWS (rather than individuals) who eventually persist or recover from stuttering. Our future research will endeavor to map the developmental trajectories of factors related to persistence in individual children to capture changes in the progression of stuttering related to the developmental stage of the child. Although the next steps in this work will contribute tremendously to our understanding of the development of stuttering, the current findings provide important information for clinicians who must make decisions about risk of persistence in 4- and 5-year-old CWS based on a snapshot of their stuttering. Unearthing additional factors associated with persistent stuttering can help clinicians and families make optimal decisions regarding intervention and allow researchers to more accurately characterize subgroups of CWS.

This study focused on speech disfluencies, the core feature of stuttering, yet we recognize that stuttering sequelae are complex and extend beyond overt speech behaviors. A comprehensive assessment would clearly include assessments of reactions from the child and parents as well as feelings and attitudes that are emerging in preschoolers who are stuttering. Nevertheless, our findings contribute to a sparse list of risk factors available to clinicians to evaluate a child's risk for persistence. Our results indicate that stuttering severity measured by the weighted SLD in 4- and 5-year-old CWS can be linked to persistence of the disorder. Measures of SLD frequency do not distinguish CWS-ePer and CWS-eRec at younger ages, thus reinforcing the idea of the dynamic unfolding of stuttering over childhood (Smith & Weber, 2017).

This study broadens the scope of the weighted SLD index to identifying CWS who may be at greater risk for stuttering persistence. We suggest that the weighted SLD be included in a comprehensive assessment with 4- and 5-year-old CWS that considers other established risk factors along with child and parent reactions to stuttering.

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Appendix

Participant Details

Subject	Status	Sex	Age (at Visit 1)	Age at onset	Duration of stuttering (at Visit 1)	Syllable count	Age at last visit/ last study visit
Participant 1	Per	m	67	30	37	1,207	115/Yr 5
Participant 2	Per	m	50	36	14	1,204	100/Yr 5
Participant 3	Per	m	48	24	24	1,129	99/Yr 5
Participant 4	Per	f	49	42	7	892	97/Yr 5
Participant 5	Per	m	58	36	22	1,046	84/Yr 3
Participant 6	Per	m	71	42	29	1,115	98/Yr 3
Participant 7	Per	m	61	48	13	1,104	98/Yr 4
Participant 8	Per	f	52	24	28	1,193	89/Yr 4
Participant 9	Per	m	60	36	24	264	85/Yr 3
Participant 10	Per	m	69	42	27	1,193	107/Yr 4
Participant 11	Per	m	65	60	5	1,200	79/Yr 2
Participant 12	Per	m	48	30	18	1,157	78/Yr 3
Participant 13	Per	f	59	36	23	1,205	98/Yr 4
Participant 14	Per	m	48	24	24	1,208	86/Yr 4
Participant 15	Per	m	68	36	32	1,206	105/Yr 4
Participant 16	Per	m	63	48	15	1,205	105/Yr 4
Participant 17	Per	f	59	36	23	1,203	94/Yr 4
Participant 18	Per	m	56	24	32	1,116	99/Yr 4
Participant 19	Rec	m	49	30	19	1,201	73/Yr 3
Participant 20	Rec	m	66	30	36	1,210	103/Yr 4
Participant 21	Rec	f	49	24	25	1,203	100/Yr 5
Participant 22	Rec	m	49	36	13	931	74/Yr 3
Participant 23	Rec	m	48	36	12	1089	103/Yr 5
Participant 24	Rec	m	51	36	15	1,120	75/Yr 3
Participant 25	Rec	m	55	42	13	1,200	80/Yr 3
Participant 26	Rec	m	48	36	12	1,069	100/Yr 5
Participant 27	Rec	m	61	48	13	1,204	113/Yr 5
Participant 28	Rec	m	49	30	19	1,208	74/Yr 3
Participant 29	Rec	m	55	24	31	1,219	104/Yr 5
Participant 30	Rec	m	57	36	21	1,203	106/Yr 5
Participant 31	Rec	f	50	24	26	1,193	99/Yr 5
Participant 32	Rec	m	53	24	29	1,213	91/Yr 4
Participant 33	Rec	f	52	36	16	1,212	91/Yr 4
Participant 34	Rec	m	49	30	19	1,204	88/Yr 4
Participant 35	Rec	m	67	33	34	1,206	105/Yr 4
Participant 36	Rec	m	56	24	32	1,203	106/Yr 5
Participant 37	Rec	m	54	36	18	1,212	92/Yr 4
Participant 38	Rec	f	59	36	23	1,201	84/Yr 3
Participant 39	Rec	m	58	36	22	1,171	82/Yr 4
Participant 40	Rec	m	55	48	7	1,069	84/Yr 3
Participant 41	Rec	f	48	33	15	1,151	85/Yr 4
Participant 42	Rec	m	58	46	12	635	94/Yr 4
Participant 43	Rec	f	58	46	12	1205	95/Yr 4
Participant 44	Rec	f	58	48	10	1,209	94/Yr 4
Participant 45	Rec	m	56	22	34	1,212	82/Yr 3
Participant 46	Rec	m	58	24	34	1,202	85/Yr 4
Participant 47	Rec	m	61	42	19	827	98/Yr 4

Note. All ages are reported in months. Two participants' family history was not available (n/a). Per = persist; f = female; m = male; Rec = recovered.