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Relation of Motor, Linguistic and Temperament Factors in Epidemiologic Subtypes of Persistent and Recovered Stuttering: Initial Findings

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1. Introduction

References to stuttering as a “complex” or “multifaceted” disorder abound in the scientific literature, reflecting a general recognition that a simple characterization of stuttering is not tenable. Although speech disfluency is its cardinal feature, stuttering, as a disorder, appears to encompass more than just speech production difficulties. It is interwoven within the language, phonological, cognitive, social, emotional, and physiological domains, creating a marked heterogeneity that is especially apparent when stuttering persists. It is fitting to point out that more than 50 years ago St. Onge and Calvert (1964) asked: “What are we studying when we study stuttering? Whatever it is, is it one, several, or many?” (p.160). While there has been a generally accepted distinction between developmental stuttering and acquired stuttering, within developmental stuttering there is no formal recognition of subtypes[1]. In this article we echo St. Onge and Calvert’s (1964) question by reporting on progress from direct testing of whether persistent and recovered stuttering are viable subtypes.

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[1]The term “subtypes” rather than “subgroups” is employed here. Subgroups can be formed for different purposes. For example, division into arbitrary subgroups may be based upon age, or assignment to a task. Other usages include statements such as “only a small subgroup of participants responded…” In contrast, clinical subtypes entail assumed or observed consistent differences in symptomatology and/or etiological factors.

Historically, there have been proposals for subtype classification of stuttering reflecting diverse orientations. Some were based on presumed etiology (Brill, 1923; Canter, 1971), some on different phenomena of stuttering (Douglas & Quarrington, 1952; Froeschels, 1943; Schwartz & Conture, 1988), and others on the presence or absence of concomitant disorders (Blood & Seider, 1981; Riley, 1971). Biological differences have also been linked to possible subtypes (Hinkle, 1971; Poulos & Webster, 1991). Van Riper (1971) attempted to differentiate stuttering based on distinct developmental courses of the disorder. Furthermore, these classifications ranged from being based on a single domain, such as different psychological states (Brill, 1923) to multiple domains, such as St. Onge's (1963) triple types: psychogenetic, organic, and speech symptoms (Yairi, 2007).

The various subtype proposals, however, have been accompanied by little research or convincing evidence. In an early study, Berlin (1954) compared 110 people who stutter (PWS) divided into seven apriori defined subtypes based on: (a) family history, (b) laterality, (c) home environment, (d) presumptive brain damage, (e) diadochokinesis, (f) maladjustment, and (g) dysphemia. They were examined in relation to 12 variables including stuttering onset, disfluency, personality, diadochokinesis, and health history. The only significant findings were that presumed brain damage was associated with a more gradual onset and higher scores on the Minnesota Multiphasic Personality Index (MMPI). Hinkle (1971) pioneered the research of subtyping in relation to brain structure and function reporting that PWS who differed in brain lateralization during dichotic listening also differed in their stuttering patterns, severity, and level of the adaptation effect. Kroll (1976) reported high accuracy in parting interiorized from exteriorized stuttering, whereas Schwartz and Conture's (1988) cluster analyses of speech samples yielded a distinction between PWS predominantly exhibiting repetitions and those predominantly exhibiting sound prolongations. Feinberg, Griffin and Levey (2000) reported that subtypes could be discerned along personality, cognitive, and intellectual dimensions.

A number of recent brain studies of PWS have explored within-population differences. For example, atypically large right planum temporale in PWS was associated with greater disfluency than that of PWS with more typical morphology (Foundas, Bollich, Feldman, Corey, Hurley, & Heilman, 2004a; Foundas, Corey, Hurley, & Heilman, 2004b). They also responded differently to altered auditory feedback (AAF), a finding reminiscent of Hinkle (1971). The first brain structure study of children who stutter (Chang, Erickson, Ambrose, Hasegawa-Johnson & Ludlow, 2008), conducted at the University of Illinois with 9- to 12-year-olds, revealed differences in fractional anisotropy, a measure of white matter integrity, between children who persisted in, and those who recovered from, stuttering. The latter exhibited poorer integrity of fibers connecting mostly left cortical centers. Most recently, Chang, Zhu, Choo, and Angstadt (2015) employing a larger sample of considerably younger children (down to age 3), also found that the level of fractional anisotropy in tracts interconnecting auditory-motor areas and tracts that support skilled movement control differentiated CWS. Those with low fractional anisotropy had more severe stuttering than those with high fractional anisotropy. Additionally, there were statistically significant sex differences among CWS in the patterns of white matter development.

The Illinois longitudinal studies (see a comprehensive summary in Yairi & Ambrose, 2005) contributed considerable evidence for subtypes based on diverging developmental paths during the first few years after the disorder's onset. Our findings indicate two broad categories of developmental stuttering: (1) persistent, lasting more than 3 or 4 years after onset, and (2) natural recovery, showing complete remission within 3 to 4 years following onset. The recovery process can be seen during the first year of stuttering, although the process for the majority of cases tends to be completed during the second and third year post onset. There is not a continuous distribution of cases as the incidence of recovery drops sharply after that.

Our longitudinal measures of observable stuttering have been reinforced by segregation analyses on the pedigrees of 66 young CWS that provided positive evidence for genetic differences between persistency and recovery (Ambrose, Cox, & Yairi, 1997). Our team's genotyping studies also yielded persistent-recovered differences in chromosomes on which genes underlying stuttering were suspected to be located (Suresh, Ambrose, Roe, Pluzhnikov, Wittke-Thompson et al., 2006; Wittke-Thompson, Ambrose, Yairi, Roe, Ober, & Cox, 2007).

Other distinctions supporting persistence and recovery subtypes were also reported for language and phonology (Yairi & Ambrose, 2005; Watkins & Yairi, 1997). For example, language skills of both persistent and recovered children were found to be slightly precocious near onset, but only the recovered group returned to a normative level while the persistent group continued with higher than expected skill levels. In phonology, the development of the late 8 phonemes lagged in persistent children compared to recovered and control children, even though the pattern of development was within normal limits for all the children (Paden, Ambrose, & Yairi, 2002). The contribution of language to persistent-recovered subtypes has been reinforced by recent studies at the Purdue University Stuttering Project. They have reported that 6–8 year-old children who persisted in stuttering differed from those who recovered in their neural processing of linguistic information indexed by event-related brain potentials (ERPs) elicited by linguistic stimuli, despite having normal language skills. Further, brain wave patterns elicited for semantic processing may help predict persistency (Mohan, Hampton Wray, & Weber-Fox, in press; Usler & Weber-Fox, in press).

Evidence that motor deficits are also a potential factor in persistent-recovered differentiation was also reported by the Purdue team. Olander, Smith, and Zelaznik (2010) showed that children who persisted in stuttering exhibited higher motor coordination variability than their recovered peers. Furthermore, for those who recovered, coordination scores were back to normal with the passage of time. A few years later, Spencer and Weber-Fox (2014) reported that preschool speech articulation and nonword repetition abilities, both involving motoric functioning, may also help predict eventual recovery or persistence of stuttering.

So far, temperament studies comparing persistent with recovered children by means of formal instruments have not been published. In their analyses of responses of parents to several items pertaining to behavior problems in a case history questionnaire, Yairi and Ambrose (2005) noted that there were only slight differences, early in the course of stuttering, between children who were later classified as persistent or recovered. Temperament, however, has been actively researched in pediatric stuttering. Eggers, De Nil and Van den Bergh (2010) examined composite temperament factors in CWS and typically developing children using the Dutch version of the Children's Behavior Questionnaire (Van den Bergh & Ackx, 2003). They found that scores for the Inhibitory Control and Attentional Shifting scales in the Effortful Control factor were significantly lower for the CWS group, whereas the scores for the Anger/Frustration scale of the Negative Affectivity factor were significantly lower for the CWS group. Recent reviews of the literature on temperament (Conture, Kelly & Walden, 2013; Kefalianos, Onslow, Block, Menzies & Reilly, 2012) have revealed variability in findings with indication of an association of some temperament factors and stuttering in early childhood. Specifically, CWS were found to score lower on adaptability and attention span, and higher on negative mood. Previous studies have not linked temperament to a persistence / recovery dichotomy.

Overall, it would appear that most past stuttering subtype classifications have been limited in some way, such as having a narrow focus not clearly situated within an articulated theoretical framework, and lacking sufficient empirical support. For a comprehensive review of the stuttering subtype literature, see Seery, Watkins, Mangelsdorf, and Shigeto (2007) and Yairi (2007). Still, recent findings summarized above are encouraging. Although no current stuttering theory accounts for subtypes of persistence and recovery, their possible reality holds theoretical, research, and clinical implications for the disorder of stuttering in general. First, research designs pertaining to childhood stuttering would undergo major upgrading in terms of accurate and meaningful participant selection, reducing the inconsistency in reported findings for many variables in children who stutter. Second, and equally important, is the compelling need to better understand clinical variants to facilitate diagnostic and prognostic accuracy, as well as treatment efficacy. Stuttering subtypes information will enhance risk/non-risk prediction for children who begin stuttering, allowing a focus of resources on early intervention for the children at greatest risk of chronicity (Yairi, 2007).

Hence, based on the above review, our most recent longitudinal research program has focused on persistence – recovery comparisons in reference to the general domains of stuttering epidemiology, language, motor control, and temperament because they often differentiate stuttering from the speech of normally fluent children albeit with heterogeneous and complex findings. No-one has previously followed these four areas in the same group of stuttering participants for several years following onset. Our hypothesis based on ours and others' work is that persistency and recovery are identifiable subtypes not only as defined a priori in terms of fluency but with characteristics that extend across epidemiology, motor control, language, and temperament domains. We posit that our findings will show that recovered CWS bear more similarity to typically developing children, whereas persistent CWS differ from either group. This report summarizes the central findings of a multi-center longitudinal study of stuttering aimed at exploring subtypes of persistence and recovery by profiling epidemiology, language, motor, and temperament variables.

2. Method

2.1. Participants

Participants were identified at four data collection sites (University of Illinois at Urbana-Champaign, University of Iowa, Northern Illinois University, and University of Wisconsin at Milwaukee), applying identical criteria. All children gave verbal or implied consent and parents/guardians gave written consent for themselves and for their children. All procedures were approved by the IRB at each institution. Whenever available, the parents participated in several ways by providing case history, progress report information, participating in speech samples, and filling out temperament questionnaires.

Stuttering Group—Eighty-one preschool age children who stutter (CWS) were recruited from the respective Midwest university communities and their surrounding areas. They were identified through a recruiting campaign involving daycare centers, speech-language clinicians, health professionals, advertisements and self-referrals. Such a range of sources results in a reasonably representative sample of the population of young children who are beginning to stutter. To assure epidemiological consistency, the sample only included children within 12 months of stuttering onset before the first visit. This excludes children who have already undergone undocumented changes (including early recovery) that may affect the course of stuttering, and minimizes weighing the sample toward persistent stuttering.

All stuttering preschool participants met the following entry criteria: (a) parental judgment that stuttering is present, (b) speech-language clinician’s (certified staff member) judgment of stuttering, (c) parental rating of stuttering severity of >= 1 on an eight point scale stuttering severity scale ranging from zero as “no stuttering” to 7 as “very severe stuttering,” (d) clinician rating of stuttering severity of >= 1 on a similar eight-point scale, (e) minimum frequency of stuttering-like disfluencies (SLD, detailed in the next section) of 3.00 per 100 syllables, and (f) within 12 months of the reported onset of stuttering. Children with documented evidence of neurological disorders were excluded.

Because children entered the study at different times during their first year of stuttering, and because some did not continue the study for the full possible 5 years, the period of observation varied. Eleven children were observed for 1–2 years following onset, 17 were observed 2–3 years, 30 for 3–4 years, and 23 for 4–5 years following onset. As would be expected, there were more boys than girls in the stuttering group.

Upon completion of the study, children were classified as Persistent or Recovered when data permitted. With the Control group (described below), this allowed for three-way comparisons. Classification criteria followed those established by Yairi and Ambrose (2005). For persistency they were: (a) more than 3 stuttering-like disfluencies (SLD) per 100 syllables, (b) rating greater than or equal to 1 on a 0–7 stuttering severity scale assigned by investigator or parent, and (c) stuttering observed and/or reported by parent or child for at least 42 months. Criteria for recovery were: (a) fewer than 3 SLD per 100 syllables, (b) rating lower than 1 on the 0–7 stuttering severity scale assigned by parent(s) and investigator independently, and (c) stuttering present at entry to study but at some later point absent for a continuous period of at least 12 months (and through end of participation in the study).

Of the 81 children who stuttered, 58 were eventually classified based upon data gathered over the four years of the study: 19 as persistent and 39 as recovered, indicating a recovery rate of 67%. The remaining 23 CWS (17 males, 6 females) were not determined to be persistent or recovered because they did not complete the study, leaving before persistence or recovery could be documented. Data are reported only for the classified children.

Control Group—Forty normally fluent control children (NFC) also participated for control purposes. They met the following criteria: (a) parental report of a negative history of stuttering, (b) investigator judgment that the child had not ever exhibited stuttering, (c) rating lower than 1 on the 0–7 stuttering severity scale assigned by parent(s) and investigator independently and (d) a negative history of neurological disorders. These children were recruited primarily through advertisement in the same geographical area as the stuttering children.

There were no statistically significant differences in maternal education among the three groups. Age, gender distribution, and fluency measures are presented in Table 1.

2.2. Procedures

The longitudinal study was set up as a series of 7 visits. The first visit (Visit 1) was within 12 months of stuttering onset. The follow-up visits were set up according to the time period following Visit 1: Visit 2, 6 months later, Visit 3, 1 year later, Visit 4, 18 months later, Visit 5, 2 years later, Visit 6, 3 years later and Visit 7, 4 years later. The data reported here derive from Visits 1, 3, and 5 – corresponding to entry to the study as well as one and two years later. Disfluency data for Visit 6 are also reported. The four regional data collection sites performed a full set of identical assessment procedures in four domains: Epidemiology, Motor, Language, and Temperament. Only a subset of the data obtained for each domain is presented in this initial report.

2.2.1. Epidemiology

Parent interview: The initial evaluation included an in-depth interview with one or both parents. Supplementary information was solicited, when appropriate, from an absent biological parent or grandparents. A standard coded questionnaire pertaining to (a) family background, (b) health and developmental history along with, (c) date, manner (gradual-sudden), and circumstances of stuttering onset and (d) characteristics of the child's very early stuttering (see Yairi and Ambrose, 2005) was completed. Parents also rated stuttering severity using the 8-point (0–7) severity scale described above. They were asked to place their child on this scale before any feedback from clinicians.

A clinician severity rating (Ambrose & Yairi, 1999; Yairi & Ambrose, 1999, 2005) was also obtained. In this scale, scoring is broken down into 4 components: frequency of SLD, duration, tension, and accessory characteristics. The first three are rated from 0 to 6 and their mean calculated. Accessory characteristics were rated from 0 to 1, and this number is added to the mean of the first three items. A maximum score of 7 ("very severe" stuttering) could thus be obtained if frequency, duration, and tension were rated as 6 and accessory characteristics as 1.

Speech samples: Audio and video recordings of the children's speech were obtained in a sound-treated room suited for young children. Forty-minute audio and video recordings of speech samples (20 minutes on each of two days) were made with a Shure omnidirectional microphone (MX393/O), connected to a Mackie 1202-VLZ mixer, and a Panasonic WV-CL830 color TV camera. The microphone was positioned on an elevated plastic surface 15 inches directly in front of the child's mouth. A lapel microphone was also attached to the child’s clothing approximately 6 inches from the child’s mouth. The audio and video signals were recorded onto DVD, VHS, and mini-DV, with an additional CD audio recording.

Each recording/testing session consisted of two visits separated by up to a week to achieve better representation of the child’s speech, administer all tests and examinations, obtain parent progress reports, and minimize the effects of mood, conversational topic, and common fluctuations in stuttering. Approximately 50% of the speech sample was recorded during verbal interaction with the parent(s) and 50% with one of the experimenters. The 40 minute-recording allowed for speech samples of 1,000+ syllables. Samples were analyzed to identify and tally three types of disfluency: part-word repetitions (PW), single syllable word repetitions (SS), and disrhythmic phonation (DP, blocks and prolongations). The frequency of each type per 100 syllables was determined for each participant for each visit. The sum of the three disfluency types constitutes our measure of stuttering-like disfluencies (SLD). Also, the mean number of Repetition Units (RU), the number of extra productions (prior to the final) of either syllables or single-syllable word repetitions was calculated for each participant. Disfluencies were initially marked by a trained research assistant. All samples were checked in their entirety by a second trained research assistant. One of the investigators (NA) provided assistance when needed. Any discrepancies were resolved by consensus.

2.2.2. Motor

Speech kinematics: Jaw displacement was measured in a subset of children in Visits 1, 3, and 5, using a protocol developed by Moon and Zebrowski (2002). This procedure required the child to sit relatively still wearing a football-type helmet. The speech stimuli consisted of three increasingly complex utterances: “papa,” (simple) “buy papa,” (moderate) and “buy papa a puppy” (complex). The most complex sentence was intended to be a motoric challenge, especially at the first visit when children were youngest. After an utterance was modeled by the experimenter, the child immediately repeated it. Fifteen consecutive tokens were obtained at a comfortable rate before progressing to the next level of utterance complexity. For each participant, ten perceptibly fluent productions from the 15 of the complex level were selected for analysis. Jaw movements were recorded via a strain-gauge transducer mounted on a cantilever beam whose distal end inserted into small plastic tube taped securely to the child’s chin. The proximal end of the transducer was inserted into a bracket on the helmet. The helmet, chosen to match the head dimensions of small children, stabilized the head so that jaw movements could be made independently from head motion. The single transducer was balanced to pick up the opening-closing dimension of jaw motion (calibration: ±1 mm). The transducer signal was amplified with a Biocommunication amplifier (amplification – 10x; low pass filter – 50 Hz) and digitally sampled 5,000 times per second with a Windaq A/D board and Windaq/Pro software (DATAQ Instruments, Inc., Akron, OH). Acoustic recordings were made with a Shure omnidirectional lapel microphone (MX393/O) worn by the participant. It was amplified via a Mackie 1202-VLZ mixer and then similarly sampled on a second channel of the Windaq A/D board. The acoustic recording was solely for reference and was not analyzed. Because a number of the children were uncomfortable with the helmet and transducer, the subject sample for the kinematic measures is smaller than for the other measures and not consistent across visits.

The Windaq data were imported into Matlab (ver. R2010A, Natick, MA), downsampled to 100 samples/sec and low pass filtered at 15 Hz (Butterworth 4 pole digital filter). Custom routines were then applied to the jaw displacement signal to derive velocity profiles. The displayed profiles revealed points of peak velocity in the signal used as referents for measurement.

The variability of jaw displacement for an utterance was measured with the spatiotemporal index (STI, Smith, Goffman, Zelaznik, Ying, & McGillem, 1995). The portion of the jaw displacement signal used for the STI calculation included points between the peak velocity of the first downward motion of the jaw in the first syllable of ‘buy’ and the last upward jaw motion in the second to the last syllable ‘pup’. These points were labeled with an automated routine. Each trial was then normalized in terms of displacement (Z score) and interpolated onto a common time base of 1000 points following Smith et al., 1995. The standard deviation of normalized displacement was then sampled at 50 time points across the 10 utterances and summed to generate the STI. The duration of kinematic motion (henceforth referred to as “kinematic duration”) between these points was also recorded for each trial (prior to temporal normalization).

Fundamental frequency (F_0) variability: The variation in vocal F_0 was investigated in VCV contexts by measuring cycle-to-cycle variation preceding and following a voiceless stop based closely on the methods of Robb and Smith (2002) and Arenas, Zebrowski and Moon (2012). The variability of fundamental frequency (F_0) change associated with devoicing and onset of fundamental frequency in the VCV sequences /apa/ and /itu/ was then compared across the groups and visits (1, 3 & 5). The participants repeated utterances at three levels of complexity: “papa” and “see two” (simple), “I see papa (verb)ing” and “I see two (noun)s” (moderate) and “I see two papas” (complex) in separate trials. The cues to produce the utterances were pictures and verbal prompts provided by an investigator. At least 6 utterances had to be produced correctly in order for a participant to be included in this analysis. The productions were recorded by a Shure omnidirectional microphone (MX393/O) connected to a Mackie 1202-VLZ mixer. Participants were not wearing the kinematic apparatus during these recordings. The speech stimuli were digitized at a 10,000 Hz sampling rate (16 bit resolution) using Computerized Speech Lab (Kay CSL-4300B series).

For analysis, tokens of the /apa/ and /itu/ VCV syllables were displayed as amplitude-by-time waveforms and the vocalic portions were demarcated by two vertically oriented cursors and then extracted for F_0 estimation. Only syllables with perceptually identifiable vowels were included. The variability of F_0 offset and F_0 onset were measured from each waveform as follows:

F_0 offset (devoicing) variability was measured by examining the change in period across the last 10 vocal cycles of the vowel (i.e. /a/) preceding the voiceless stop consonant /p/ or /t/. The negative peaks of these cycles of the vowel were marked in each waveform using CSL software. F_0 for each period was determined by calculating its reciprocal. To minimize the effects of within and between subject variability, each of the 10 vocal cycles were normalized by converting to semitone values following Arenas et al. (2012). Similar to F_0 offset, F_0 voicing onset variability was measured by marking the first 10 negative peaks following vocalization onset after the voiceless stop consonant. The intervals or period between the peaks across the first 10 vocal cycles were converted to F_0 and then normalized by converting to semitones.

2.2.3. Language—The language assessment included several standardized measures: (a) the *Test of Early Language Development* (TELD, Hresko, Reid, & Hammill, 1999), that assesses both expressive and receptive language proficiency against solid normative data; (b) the *Peabody Picture Vocabulary Test-III* (PPVT, Dunn & Dunn, 1997), commonly used to assess receptive vocabulary and having strong psychometric foundations; and (c) the *Expressive Vocabulary Test* (EVT, Williams, 1997), a counterpart to the *PPVT-III*. MLU and MLU-Z scores were calculated (Leadholm & Miller, 1992).

Phonological skills were assessed by *Percent of Consonants Correct* (PCC, Shriberg & Kwiatkowski, 1982). Ten to 15 minutes of the conversational language samples, including at least 90 different words, were evaluated. Samples were transcribed following a system of narrow-phonetic transcription and conventions developed for research in child phonology (Shriberg & Kent, 1982; Shriberg, Kwiatkowski, & Hoffman, 1984), and were formatted for computer analysis using the PEPPER system (Shriberg 1986, 1993). The program provided the *Percentage of Consonant Correct* (PCC, Shriberg & Kwiatkowski, 1982). PCC is a measure of the percentage of intended consonants produced correctly, with all deletions, substitutions, and clinical distortions counted as incorrect. Results were evaluated separately for eight early developing sounds (Early-8), eight middle developing sounds (Middle-8) and eight late developing sounds (Late-8) as described by Shriberg (1993). Inter-rater reliability was calculated on 10% of samples using an agreement analysis. Overall agreement on consonant transcription was 80.8% for narrow transcription and 92.3% for broad transcription.

2.2.4. Temperament—Children's temperament was measured with the *Children's Behavior Questionnaire Short Form Version I* (CBQ, Rothbart, Ahadi, Hershey, & Fisher, 2001; Putnam & Rothbart, 2006) that was administered to both parents whenever possible. The short form was used to increase parent compliance in completion of the questionnaire. The CBQ is a well-normed instrument with high validity that has been successfully used in other research on temperament and childhood stuttering (Eggers, De Nil & Van den Bergh, 2010). Its short version consists of 94 items scored in the following manner: 1 =Extremely Untrue, 2 = Quite Untrue, 3 = Slightly Untrue, 4 = Neither True or Untrue, 5 =Slightly True, 6 = Quite True, 7 = Extremely True, with a Not Applicable (N/A) option available. The scale rates the child on 15 different behavior dimensions that combine to form three composite scores: (a) surgency/extraversion (activity level, approachability, high intensity pleasure, impulsivity, and shyness), (b) negative affectivity (anger/frustration, discomfort, fear, sadness, and soothability), and (c) effortful control (attentional focusing, inhibitory control, low intensity pleasure, perceptual sensitivity, smiling and laughter).

2.3. Data Analyses

The data were analyzed using SPSS v.22 analysis of variance for each domain, for each of the three visits, with group (persistent, recovered, control) as the independent variable and measures within each domain as the dependent variables. For variables that reached significance (p=<.05), post-hoc Bonferroni tests were used to distinguish between the levels (p=<.05). Sample sizes and effect sizes are given for each analysis.

3. Results

3.1. Epidemiology

3.1.1. Disfluencies—The mean frequency of SLD (per 100 syllables) for persistent, recovered, and control groups is shown in Table 2, including the three SLD components, part-word repetitions (PW), single syllable word repetition (SS), disrhythmic phonation (DP, blocks and prolongations), as well as repetition units (number of times a PW or SS is repeated). Disfluencies are reported for Visits 1, 3, 5, and 6.

At Visit 1, the number of SLD for persistent and recovered groups is similar. By Visit 3, one year later, the persistent group has a greater number of SLD than the recovered, who have a greater number than the control group. At Visits 5 and 6, the persistent group (by definition) continues to stutter but the recovered and control groups appear indistinguishable. We created Z-scores based on the distribution of disfluencies in the speech of normally fluent children by combining data for all of the Illinois studies prior to the present study. Because SLD is simply a composite measure of PW, SS, and DP, Z-scores were created for the individual disfluency types only. Means for typically fluent 2-, 3-, 4- and 5-year olds did not differ statistically and so were combined. The means and standard deviations (in parentheses) for stuttering-like disfluencies of this normally fluent group of 105 children were: PW 0.57 (0.43), SS 0.63 (0.49), DP 0.18 (0.22), RU 1.09 (0.10).

Using these values, Z-scores were calculated for children in the current study for Visits 1, 3, and 5 and are shown in Figure 1. MANOVAs (one for each visit) revealed statistically significant differences between groups for all disfluency types for all three visits. Analysis of variance and post-hoc test values are given in Appendix A. As illustrated in Figure 1, the pattern of statistically significant differences for all four disfluency measures was consistent with three minor exceptions. Persistent vs. recovered differences were not statistically significant at Visit 1, but were significant at Visits 3 and 5, excepting repetition units (RU) at Visit 3. At all three visits, persistent vs. control comparisons showed significant differences for all disfluencies. Recovered vs. control differences were significantly different at Visit 1, but not at Visit 5. At Visit 3, where the Recovered group’s disfluencies are decreasing, there were significant differences for SS and RU, but not for PW or DP.

In summary, at the first visit, both stuttering groups differ from the control group; at Visit 3, persistent and control remain different but recovered is intermediary; and at Visit 5, the persistent group can be distinguished from the recovered and control groups.

3.1.2. Sex Distribution—The male to female ratios for persistent and recovered groups are shown in Table 3. A chi square test revealed no statistically significant difference between the ratios for the two groups.

3.1.3. Age at Onset of Stuttering—Overall, for the stuttering group, the mean age at onset was 35.14 months (sd = 8.97), range 19–68 months, with 58% of the onsets occurring by age 36 months and 84% by age 42 months. For the persistent group, the mean age at onset was 36.95 months (sd= 12.08) and 34.26 months (sd= 7.00) for the recovered group. The difference was not statistically significant.

3.2. Motor

Speech kinematics—Sample sizes for Visit 1 were Persistent=1, Recovered=4, Control=6; for Visit 3, Persistent=2, Recovered=7, Control=11; and for Visit 5, Persistent=7, Recovered=14, Control=10. The STI findings are shown in Figure 2. ANOVAs for each visit indicated a significant effect only at Visit 5 ($F(2,28)=7.32$, $p=.003$, $\eta_p^2=.34$) with the Recovered group displaying greater kinematic variability than the Control group ($p=.002$).

Fundamental *frequency* (F_0) variability—For the acoustic analysis, sample sizes for Visit 1 were Persistent=9, Recovered= 22, and Control=18; for Visit 3, Persistent=14, Recovered=34 and Control=24; and for Visit 5, Persistent=7, Recovered=17 and Control=9. There was a statistically significant effect only at Visit 3 for F_0 offset ($F(2,69)=4.82$, $p=.01$, $\eta_p^2=.12$), with the Persistent group displaying greater variability than the Control group ($p<.01$).

3.3. Language

3.3.1. Standardized Tests—Comparison of PPVT, EVT and TELD receptive (TELD-R) and expressive (TELD-E) scores revealed significant differences at each visit. The mean scores are shown in Figure 3 and given in Appendix B.

Visual inspection indicates that the persistent group consistently had lower scores on all tests for all visits. For all statistically significant comparisons, the Persistent group's score fell below that of the Recovered or Control groups' scores.

For the PPVT, there were no statistically significant differences at Visit 1. At Visit 3 there were significant differences ($F(2,95)=4.89$, $p=.01$, $\eta_p^2=.10$) between the persistent and recovered ($p=.02$) and between persistent and control ($p=.01$) as well as at Visit 5 ($F(2,94)=4.01$, p=.02, $\eta_p^2=.08$) between persistent and recovered ($p=.04$) and between persistent and control ($p=.03$).

For the EVT, a statistically significant difference occurred at Visit 1 ($F(2,95)=4.73$, $p=.01$, $\eta_p^2=.09$) between persistent and recovered ($p=.01$). There were also significant differences at Visit 3 (F(2,95)=3.68, p=.03, η_p^2=.07) between persistent and recovered (p=.05) and persistent and control (p=.04), and at Visit 5 (F(2,94)=3.26, p=.04. η_p^2=.07) between persistent and recovered (p=.04).

The TELD-R (receptive) test scores revealed statistically significant differences at Visit 3 (F(2,95)=3.42, p=.04, η_p^2=.07) between persistent and control (p=.03)and at Visit 5 (F(2,94)=5.97, p=.01, η_p^2=12) between persistent and recovered (p=.01) and between persistent vs. control (p=.01) groups. There were no significant differences in the TELD-E (expressive) test scores.

3.3.2. MLU—There were no statistically significant differences in MLU or MLU-Z scores between any pair of groups (persistent, recovered, control) at any visit. Nor were there any apparent consistent trends (other than increasing MLU with age).

3.3.3. Phonology—Phonological accuracy (PCC) was assessed for early, mid- and late developing phonemes. All children reached an 80% accuracy level or higher for early and mid-developing phonemes even at the first visit. Only data for late developing phonemes are presented and are illustrated in Figure 4. The one statistically significant difference occurred at Visit 1 (F(2,95)=4.24, p=.02, η_p^2=.09). Post-hoc testing revealed significant differences between Persistent and Control (p=.05) and between Recovered and Control (p=.04) groups, where the accuracy for Persistent and Recovered groups was lower than that for the Control group. Looking at scores smaller than or greater than 1 standard deviation from the mean at Visit 1, just under 25% of each of the persistent and recovered groups, but only 12% of the control group, had accuracy lower than 1 standard deviation below the mean. The percentages of each group with scores greater than 1sd above the mean were: persistent 5%, recovered 10%, and control 30%.

3.4. Temperament

The CBQ yields three composite scores: extraversion, negative affectivity, and effortful control. Both mothers and fathers completed the questionnaires, independently, at Visits 1, 3 and 5. For each subscale, scores of the two parents were significantly correlated at $p <=.003$ regardless of visit. In other words, both parents rated their children similarly and consistently in each area over a period of two years. From their points of view, these aspects of temperament, then, appear to be very stable. Data presented here are from mothers at Visit 1, the most significant of our time points regarding temperament development. Scores are illustrated in Figure 5.

When comparing scores for the three subscales, there were no significant differences for Extraversion or Effortful Control. Statistically significant differences, however, were indicated for Negative Affectivity, (F(2,88)=5.61, p=.005, η_p^2=.11). Mothers rated persistent children with higher scores (greater negative affectivity) than either recovered (p=.004) or control (p=.03) children.

Within the Negative Affectivity subscale, there were statistically significant differences for two components: fear (F (2,88=6.80, p=.002, η_p^2=.13) and soothability (F(2,88=8.66, p <. 001, η_p^2=.16). Means and SDs are reported in Table 4. Mothers rated children in the

Persistent group as more fearful than those in the Recovered group (p = .005) or the Control group (p= .002), and mothers rated children in the Persistent group as less soothable than those in the Recovered group (p <.001) or Control group (p=.042.)

4. Discussion

This report summarizes the findings of a multi-center longitudinal study of stuttering by providing a profile of epidemiology, language, motor, and temperament variables from a large sample of preschool children who stutter seen within one year post onset. Overall, the results present an intriguing picture that extends our current knowledge of the early development of stuttering. The direction of group differences was consistent in that the persistent group showed less advanced or less mature skills in language, greater variability in motor control (as indexed by acoustic data), and different temperament qualities than either the recovered and/or control groups. The recovered group only differed from the control group in phonology at Visit 1 and in kinematic variability at Visit 5.

A concise review of the results starting at Visit 1, closest to onset, shows the Persistent group differed from the Recovered group in having lower language test scores, while both the Persistent and Recovered groups lagged behind the Control group in acquisition of phonology. At this point, the Persistent group differed from the other groups in showing a more negative temperament. At Visit 3 (one year later), the Persistent still differed from Recovered and Control in general language performance, and the Persistent differed from the Control group in oral motor control (acoustic data). Finally, at Visit 5, the Persistent continued to differ from the Recovered and Control groups in language test scores, and the Recovered group differed from Controls in having more variable oral motor control as reflected in jaw movements.

4.1. Epidemiology

For the stuttering group, the age at onset and percent of onsets by ages 36 and 42 months are virtually identical to previous studies at the University of Illinois (e.g., Yairi & Ambrose, 2005) and are similar to findings of other recent investigations (Buck, Lees, & Cook, 2002; Mansson, 2005; Reilly, Onslow, Packman, Cini, Conway et al., 2013). The prior Illinois studies on recovery pointed to a later onset age and higher male to female ratio for the persistent group than the recovered group. The current data do not replicate those findings. Neither age at onset nor male-to-female ratio for persistent and recovered groups were statistically significantly different.

4.2. Motor

The STI data point to differences in variability in speech motor control between recovered and non-stuttering children at visit 5, while the acoustic data indicate variability differences between persistent and control at Visit 3, but no consistent trend in motor variables was seen across visits. The higher kinematic and acoustic variability of the Persistent and Recovered groups corresponds to the recent report by Smith, Goffman, Sasisekaran, and Weber-Fox (2012) who found elevated labial kinematic variability in a larger cohort of stuttering children. This corroboration of motor speech control variability in children suggests that increased variability in stuttering is not restricted to adults who stutter (Kleinow & Smith, 2000; Smith, Sadagopan, Walsh, & Weber-Fox, 2010), but can also be identified in CWS as young as 4–6 years of age (MacPherson and Smith, 2013). Furthermore, as indicated in the review of the literature, evidence that motor deficits are a potential factor in young children's persistent-recovered differentiation was provided by Olander, Smith, and Zelaznik (2010) and by Spencer and Weber-Fox (2014). Additional large scale studies in this direction are warranted.

4.3. Language

The current results provide a profile of language development at three time points. The data derived from the standardized test scores provided an unexpectedly consistent indication that persistent children, though within normal limits, are slightly behind the other groups on broad measures of language development. This results contrasts with Watkins and Yairi (1997) who reported that children who persist in stuttering are more likely to exhibit unusual language developmental patterns, although not necessarily low skills, than children who recover. It is common knowledge that standardized test scores cannot be used to infer fine differences in language abilities, especially because any given test samples a spectrum of language abilities; yet, consistent group differences suggest the results are tapping into underlying subtle differences in linguistic competency.

The current results do not corroborate past findings that MLU scores for both the persistent and recovered groups were higher close to onset and then remained higher for the persistent group (Watkins, 2005; Watkins, Yairi & Ambrose, 1999). Watkins (2005) even suggested that advanced language skill may be a risk factor for persistent stuttering. MLU group comparisons present difficulties, however, in that rapid growth in language skills and high variability in the timing of change across individuals results in variation ranges that are larger than mean scores.

Other investigators (Hage, 2001 Reilly et al., 2013) have reported different findings in that language scores of CWS, as a group (not separated into persistent and recovered) were higher than those of normally fluent peers. Adding to the diverging results in language ability are two recent reviews of the literature. On one hand, Ntourou, Conture, and Lipsey (2011) concluded that CWS scored lower than NFC on normative measures of both receptive and expressive language; however, it focused on children who were up to 8 years of age, which is several years beyond the typical age of onset. On the other hand, Nippold's review (2012) concluded a stuttering-language connection is not supported. There is, then, a confusing picture of early language abilities that requires consideration of what these language scores actually measure. As we stated above, standardized tests represent different constructs, while MLU likely reflects a different aspect of the early language domain.

Inasmuch as confusion concerning the role of language in early stuttering may be blamed on old, insufficiently sensitive instruments, most recent relevant studies, employing more sophisticated methods, do support our earlier contention (Watkins & Yairi, 1997; Yairi & Ambrose, 2005) that language is, indeed, a promising candidate for differentiating children who persist in from those who recover from stuttering. Specifically, scientists at the Purdue Stuttering Project have shown that event-related brain potentials (ERPs), elicited by linguistic (language and phonology) stimuli, may do just that although the children had normal language skills (Mohan, Hampton Wray, & Weber-Fox, in press; Usler & Weber-Fox, in press). Hence, further fine-tuned research is warranted.

In phonology, there were only statistically significant differences at Visit 1, where both Persistent and Recovered had lower accuracy for the late-developing phonemes. This agrees with earlier findings reported by our Illinois Studies (e.g., Yairi, Ambrose, Paden, & Throneburg, 1966). The results suggest that phonological development may hold potential for subtyping that can be explored with specific probes of phonological competency, especially at the very early stage of stuttering, a conclusion also arrived at in our earlier studies (Paden, Ambrose, & Yairi, 2002).

4.4. Temperament

The CBQ data resulted from mothers' reports at Visit 1, closest to the onset of stuttering. The measure appears to be quite robust, as scores for each child across visits and between mothers and fathers were highly correlated. The significant difference between persistent and recovered, and between persistent and control children, in the realm of negative affectivity, is thus potentially important. Parents of children who eventually persist in their stuttering appear to judge their children as having greater negative affectivity.

Previous studies have yielded contradictory findings and have complicated interpretations. CWS, as a whole, compared with NFC, have been found to be more sensitive and inhibited (Conture, 2001), less distractible and less adaptable to change (Anderson, Pellowski, Conture, & Kelly, 2003), as well as not significantly different (Reilly, Onslow, & Packman, 2009; Yairi & Ambrose, 2005). In addition, Reilly et al. (2013) stated that the first year of stuttering did not "harm" children's temperament. Tumanova, Zebrowski, Throneburg and Kayikci (2011) did not find any significant relation between CBQ scales and stuttering characteristics (disfluencies and articulation rate), in a subset of the same group of children presented here, but it is important to emphasize they did not examine persistent vs. recovered stuttering, or even the presence or absence of stuttering. Kraft, Ambrose and Chon (2014) did find temperament distinctions relating to symptomatology as shown by a significant inverse correlation between stuttering severity levels and Effortful Control ratings, i.e., as severity increased, EC scores decreased. Therefore, it remains possible there is a fundamental temperament difference in persistent children but whether its presence is related to the emergence of stuttering or its persistence is not adequately explored.

4.5. Summary and Conclusions

No current stuttering theory accounts for subtypes of persistence and recovery, perhaps because current relevant knowledge is too limited and some of it is very new. The results presented from this multi-center project, especially for language as well the hints for motor control difficulties, are in line with most recent findings reported by other investigators who explored these two domains. Adding to the picture the similarly very recent reports concerning brain morphology, there seems to be growing empirical support for reliable distinctions between Persistent and Recovered as subtypes of developmental stuttering. Our limited positive findings concerning temperament may also prove to be of some relevance but this domain, too, deserves more advanced, refined methodologies. Hopefully, progress in research on the genetics of stuttering will yield not only more specific subtype information but will also allow understanding of the contribution of each of the above domains.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

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Biographies

Nicoline G. Ambrose received her PhD from the University of Illinois. Her research centers on the etiology, onset and early development of stuttering, with particular reference to genetic factors underlying possible subtypes of stuttering.

Ehud Yairi (B.A., Tel Aviv University; M.A., Ph.D., University of Iowa), has contributed extensively to the research literature on stuttering with a special focus on the various aspects of the onset, development, and genetics of stuttering. He authored many scientific articles as well as two books: *Early Childhood Stuttering* (2005; with N. Ambrose), and *Stuttering: Foundations and Clinical Applications* (2010; with C. Seery).

Torrey Loucks (PhD - University of Toronto) is associate professor at the University of Illinois. His research centers on sensorimotor integration for speech production in typical development and in stuttering. He has published his work widely in neuroscience, motor control and speech-language pathology journals.

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Rebecca Throneburg is a professor at Eastern Illinois University. She was a doctoral student under the mentorship of Ehud Yairi and has published several articles on the development of childhood stuttering with Drs. Yairi and Ambrose.

Appendix

Continuing Education

1. Past research into subtypes of stuttering has revealed:
   - **a.** Evidence for any subtypes is quite poor
   - **b.** Linguistic, motoric and temperament subtypes have been identified
   - **c.** Persistence in and recovery from stuttering appear to be promising leads in identification of subtypes
   - **d.** Genetics work points towards subtyping but behavioral evidence is contradictory

   e. A consistent pattern but not enough data to substantiate the pattern

2. The identification of subtypes is of crucial importance because:

   a. Treatments potentially could be developed to target specific subtypes
   b. Previous studies have not been able to provide complimentary evidence
   c. Resources will not be used needlessly for treatment of children who will recover on their own
   d. Stuttering could potentially be prevented
   e. Identification of subtypes in children is an important step in discovery of subtypes in adults

3. One of the primary strengths of the study presented here is that:

   a. It gathered data from children from four different sites
   b. It gathered data on epidemiology, motor control, linguistic and temperament domains in the same group of children
   c. It gathered data for genetic analysis
   d. It gathered data from before the onset of stuttering
   e. Each of four different sites gathered data on one of the domains (epidemiology, motor control, language, and temperament)

4. Results of the study indicated that:

   a. There are indications of subtypes based on motor control, but not on language or temperament
   b. There are indications of the role of temperament but not motor control or linguistic factors in the development of persistence vs. recovery
   c. There are no clear indications of differences between persistent and recovered groups
   d. Children with persistent stuttering perform differently from children with recovered stuttering on motoric and linguistic, but not temperament, domains.
   e. There is a trend common across motoric, linguistic and temperament domains in that children with persistent stuttering tend to perform differently from children who recover or control children

5. Children with persistent stuttering scored:

   a. Higher than recovered and control children on parental judgment of Negative Affectivity
   b. Below the norm (greater than 1 standard deviation) in language tests
   c. Lower in kinematic variability than recovered or control children

- **d.** Lower in stuttering severity at onset than recovered children
- **e.** Very similarly to recovered children except at Visit 5

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    - **d.** Genetics work points towards subtyping but behavioral evidence is contradictory
    - **e.** A consistent pattern but not enough data to substantiate the pattern

    Answer: (c)
2. The identification of subtypes is of crucial importance because:
    - **a.** Treatments potentially could be developed to target specific subtypes
    - **b.** Previous studies have not been able to provide complimentary evidence
    - **c.** Resources will not be used needlessly for treatment of children who will recover on their own
    - **d.** Stuttering could potentially be prevented
    - **e.** Identification of subtypes in children is an important step in discovery of subtypes in adults

    Answer: (a)
3. One of the primary strengths of the study presented here is that:
    - **a.** It gathered data from children from four different sites
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    - **d.** It gathered data from before the onset of stuttering
    - **e.** Each of four different sites gathered data on one of the domains (epidemiology, motor control, language, and temperament)

    Answer: (b)
4. Results of the study indicated that:
    - **a.** There are indications of subtypes based on motor control, but not on language or temperament

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Answer: (e)

5. Children with persistent stuttering scored:

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- **d.** Lower in stuttering severity at onset than recovered children
- **e.** Very similarly to recovered children except at Visit 5

Answer: (a)

Educational Objectives

Readers will be able to describe the current state of subtypes of stuttering research, and to summarize possible contributions of epidemiologic, motoric, linguistic and temperament to such subtyping with regard to persistency and recovery.

Highlights

- Longitudinal data on early development in childhood stuttering presented
- Domains of epidemiology, motor, language, phonology and temperament examined
- Results support subtypes of persistence and recovery
- Further study is warranted

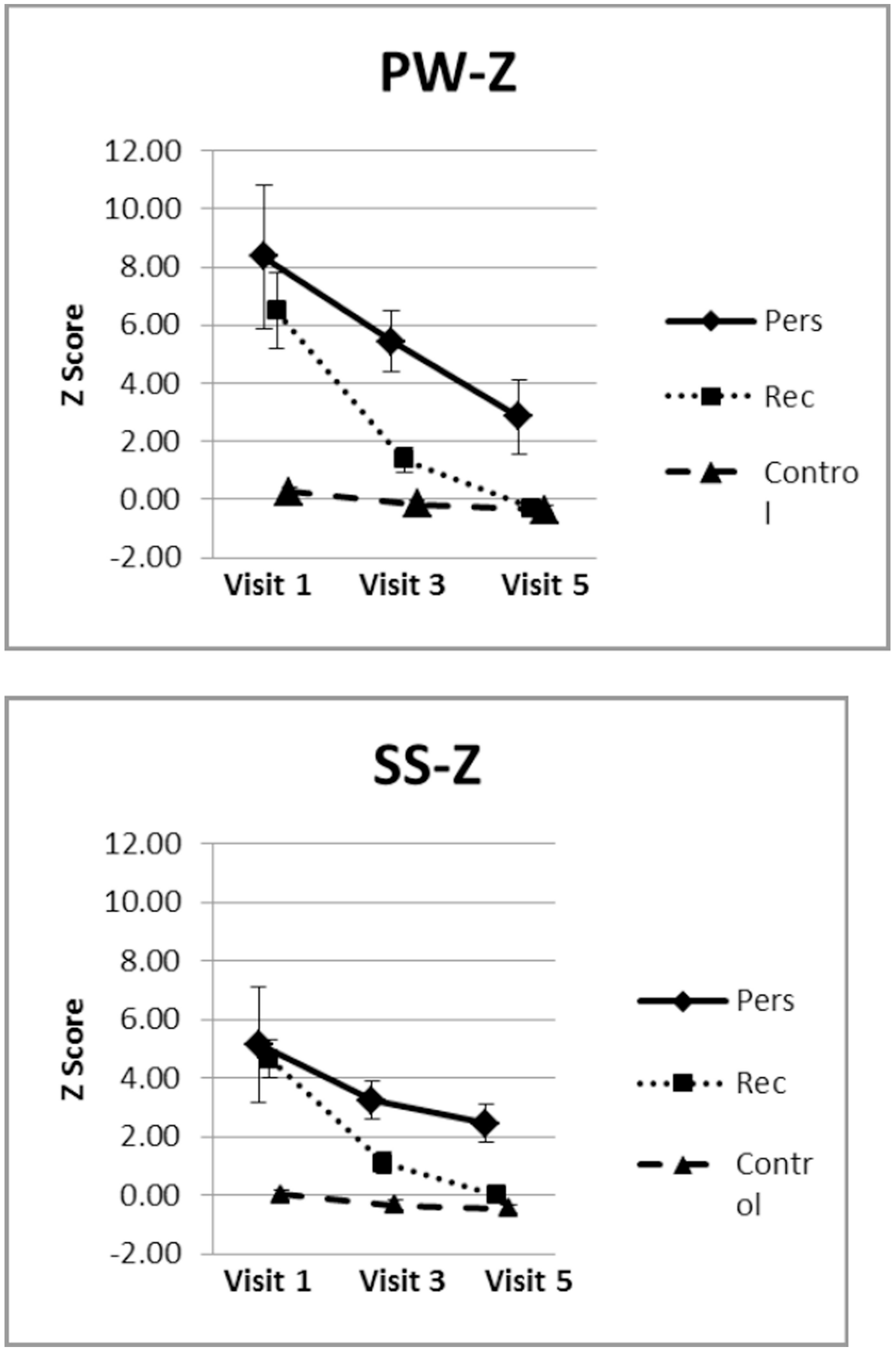

Figure 1.

Means and standard errors for disfluency measures (part-word repetitions (PW), single syllable word repetitions (SS), disrhythmic phonation (DP) and repetition units (RU) as Z-scores across visits.

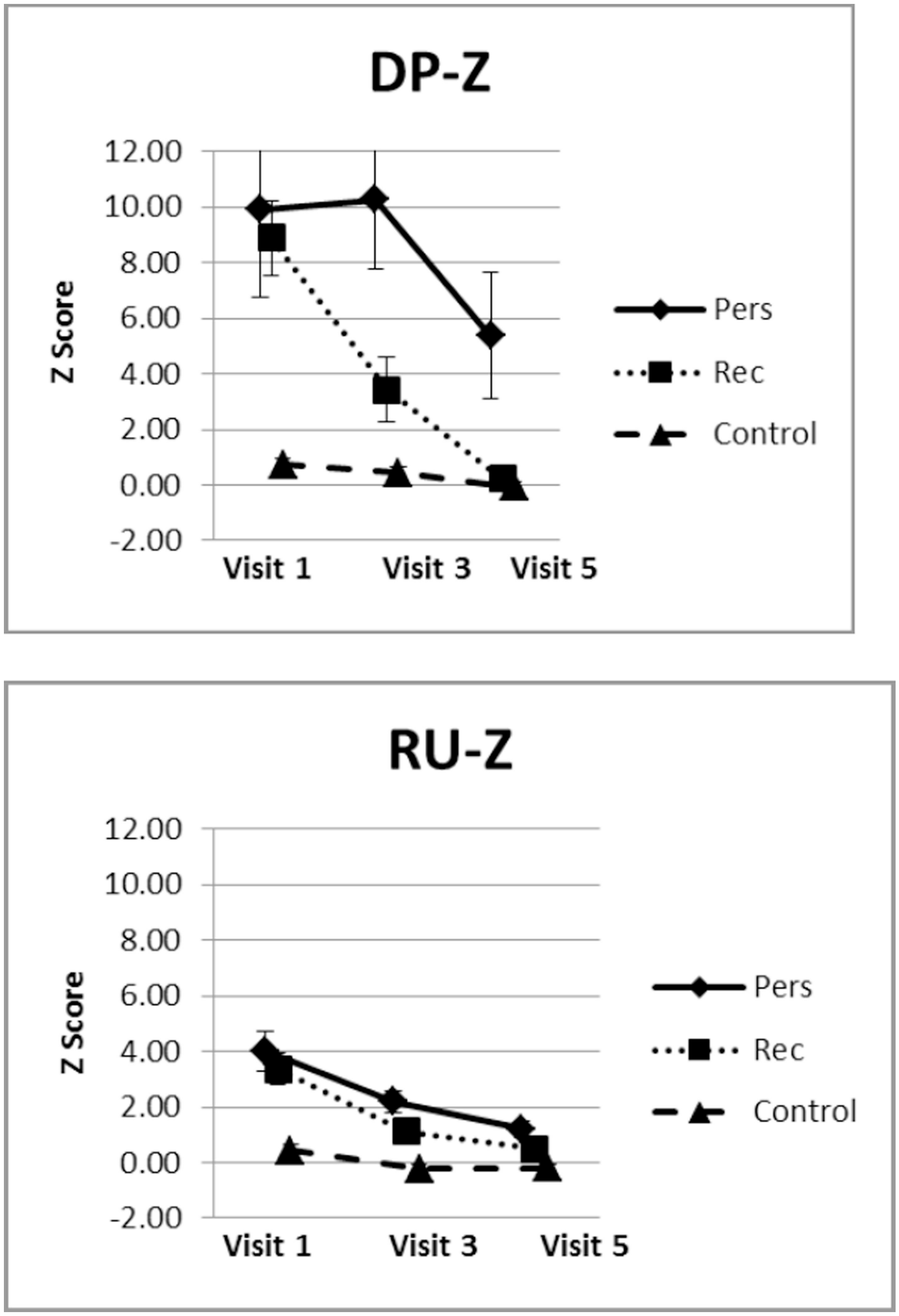

Figure 2.

Means and standard errors for spatio-temporal index (STI) and fundamental frequency onset and offset variability as standard deviation (SD) across visits.

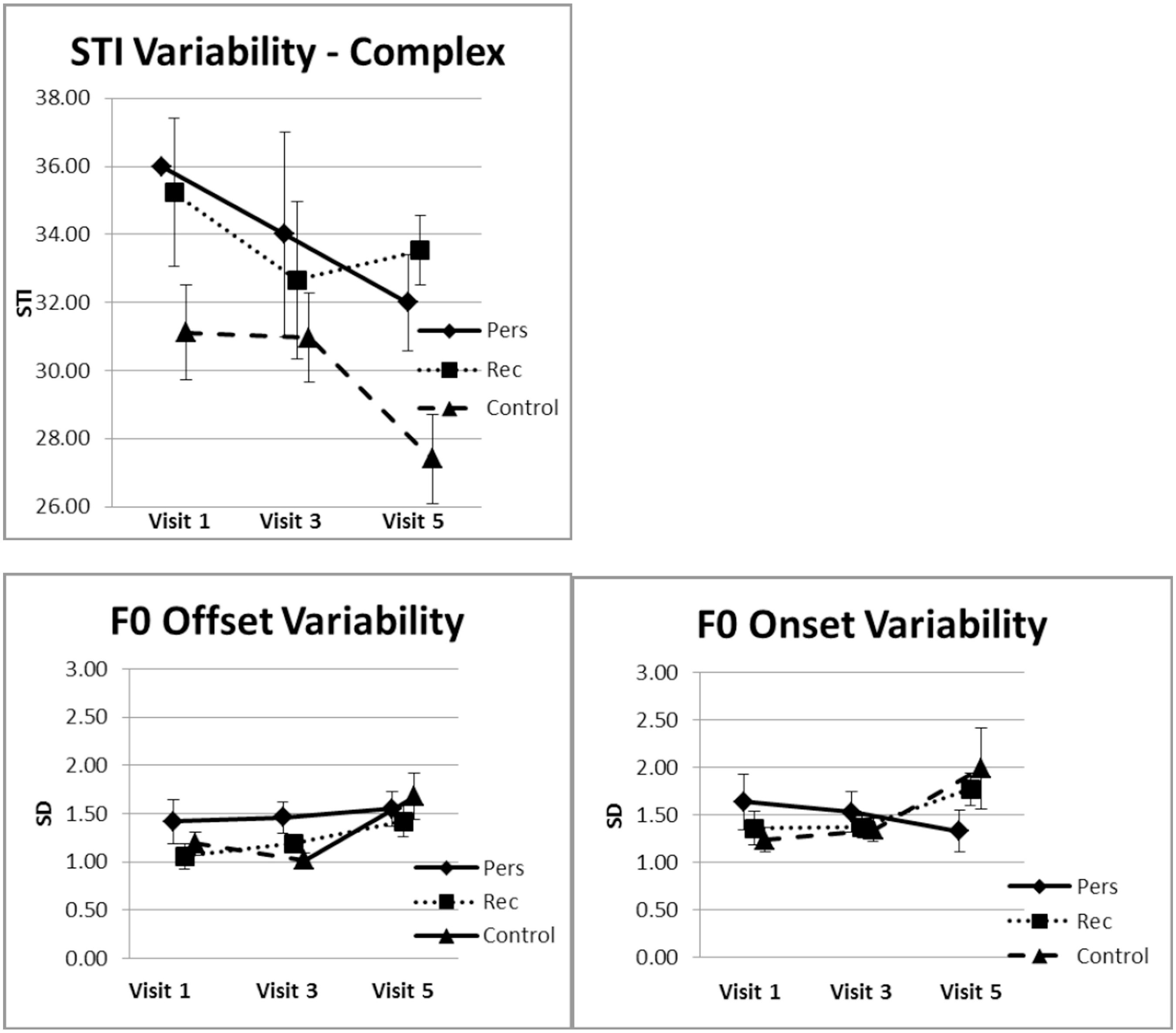

Figure 3.
Means and standard errors for standardized language test scores (Peabody Picture Vocabulary Test (PPVT), Expressive Vocabulary Test (EVT), and Test of Language Development receptive (TELD-R) and expressive (TELD-E) across visits.

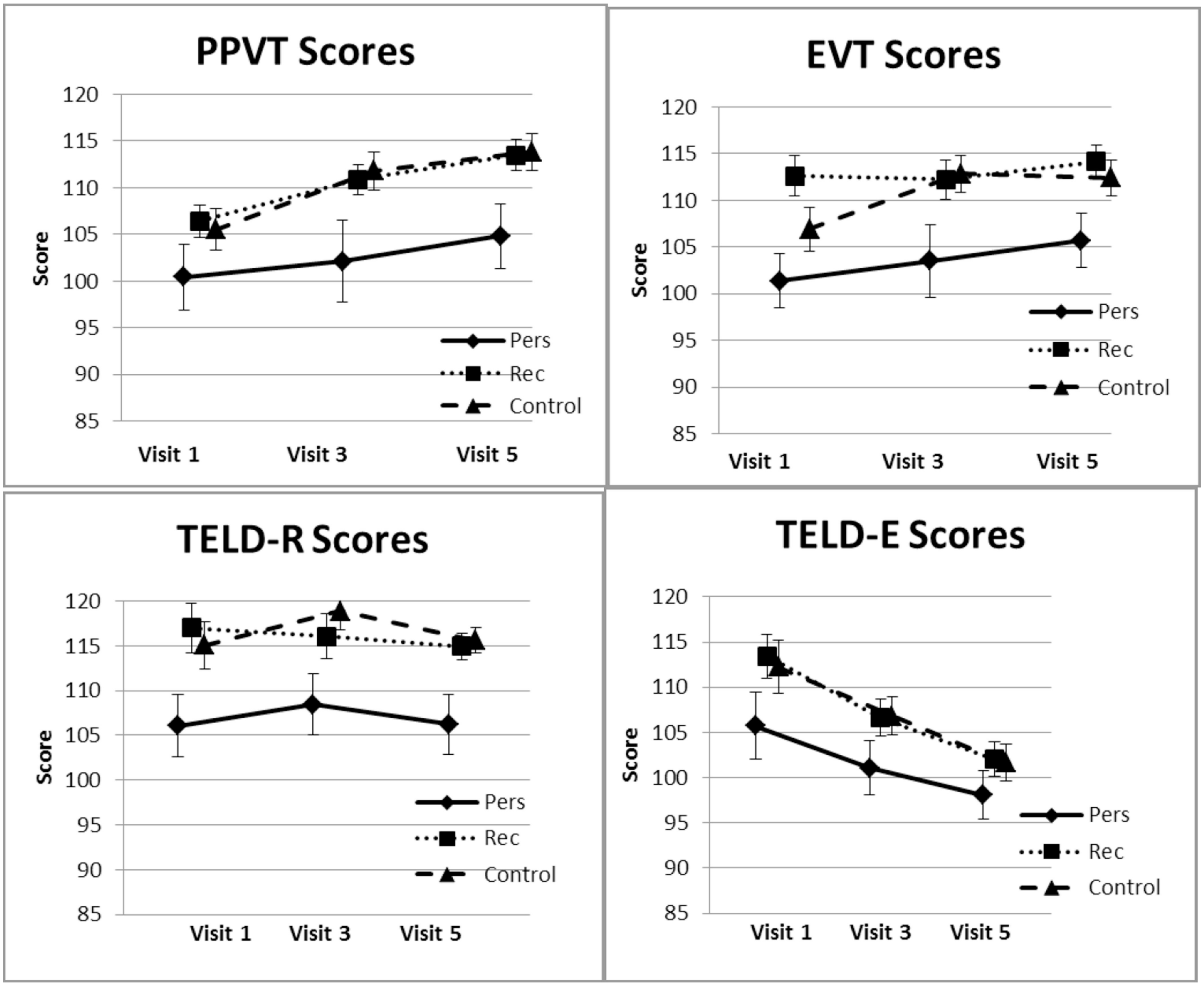

Figure 4.
Means and standard errors for phonological percent accuracy for late-developing phonemes across visits.

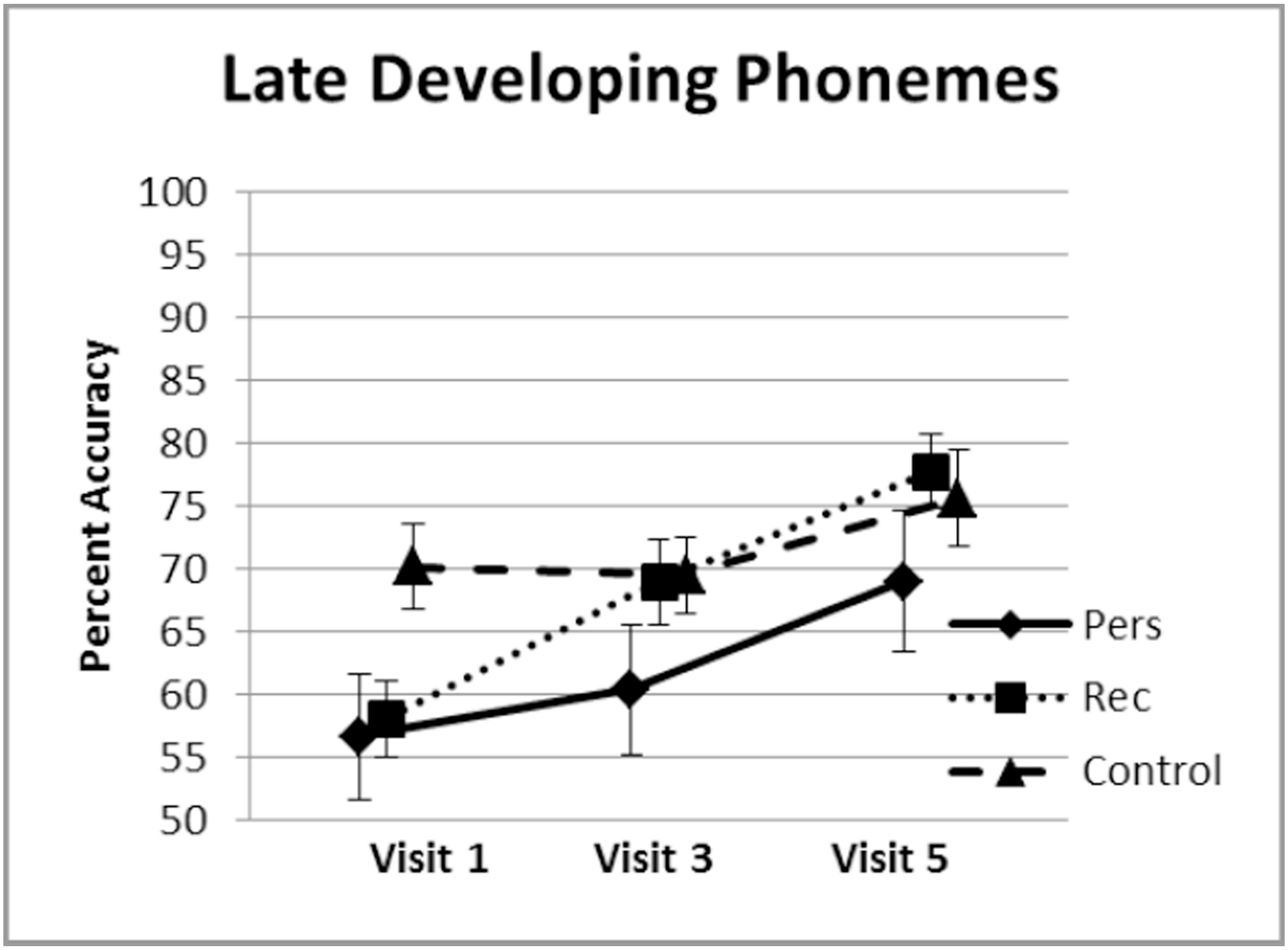

Figure 5.

Means and standard errors for Children's Behavior Questionnaire (CBQ) subscale scores across visits.

Table 1

Means and standard deviations (in parentheses) for participant groups' age, months post-onset of stuttering, and clinician and parent severity ratings at entry to the study (Visit 1).

	n	Age (months) at Entrance to Study	Months Post-Onset of Stuttering	SLD (per 100 syllables)	Clinician Severity Rating (0–7)	Parent Severity Rating (0–7)
Persistent						
Males	12	39.92 (8.36)	6.75 (4.20)	11.26 (14.19)	2.56 (1.96)	3.42 (1.49)
Females	7	50.00 (15.02)	6.57 (4.58)	6.76 (2.92)	3.22 (1.16)	4.12 (1.18)
Total	19	43.63 (11.96)	6.68 (4.22)	9.60 (11.44)	2.80 (1.70)	3.68 (1.40)
Recovered						
Males	27	37.59 (6.55)	3.63 (2.99)	9.16 (6.53)	2.67 (1.21)	3.26 (1.35)
Females	12	39.50 (9.69)	4.58 (3.73)	6.50 (4.33)	1.81 (0.91)	3.58 (0.67)
Total	39	38.18 (7.57)	3.92 (3.22)	8.34 (6.01)	2.40 (1.18)	3.36 (1.17)
Control						
Males	21	40.38 (10.15)	---	1.93 (0.92)	---	---
Females	19	44.47 (11.58)	---	1.34 (0.69)	---	---
Total	40	42.33 (10.91)	---	1.65 (0.86)	---	---

Table 2

Mean frequency and standard deviations (in parentheses) of stuttering-like disfluencies.

	Persistent			Recovered			Control		
	n	**Mean**	**SD**	**N**	**Mean**	**SD**	**n**	**Mean**	**SD**
					Visit 1				
PW1		4.16	4.68		3.36	3.54		0.69	0.40
SS1		3.06	4.04		2.85	1.91		0.62	0.45
DP1	19	2.36	3.03	39	2.13	1.81	40	0.34	0.31
RU1		1.49	0.31		1.43	0.35		1.08	0.09
SLD1		9.58	11.34		8.34	6.01		1.65	0.86
					Visit 3				
PW3		3.08	2.50		1.16	1.14		0.50	0.34
SS3		2.20	1.35		1.15	1.12		0.45	0.45
DP3	19	2.90	3.29	39	0.93	1.62	40	0.28	0.27
RU3		1.32	0.20		1.20	0.27		1.07	0.10
SLD3		8.18	6.02		3.24	3.43		1.23	0.83
					Visit 5				
PW5		2.15	2.58		0.44	0.28		0.42	0.40
SS5		1.83	1.24		0.61	0.49		0.40	0.47
DP5	18	1.49	2.02	38	0.23	0.25	38	0.16	0.28
RU5		1.23	0.14		1.14	0.29		1.06	0.10
SLD5		5.48	5.07		1.28	0.80		0.98	0.92
					Visit 6				
PW6		1.71	3.08		0.37	0.20		0.35	0.31
SS6		1.37	1.03		0.56	0.43		0.49	0.53
DP6	17	1.16	1.81	31	0.12	0.11	31	0.07	0.10
RU6		1.22	0.18		1.06	0.08		1.03	0.06

PW = Part word repetitions
SS = Single syllable word repetitions
DP = Disrhythmic phonation
RU = Repetition units

SLD = Stuttering-like disfluencies

Table 3

Male to female ratios for Persistent and Recovered groups.

	Males	**Females**	**Total**	**Male to Female Ratio**
Persistent	12	7	19	1.71
Recovered	27	12	39	2.25
Total	39	19	58	2.05

Table 4

Mean scores and standard deviations (in parentheses) for significant Negative Affectivity components.

	n	Fear	Soothability
Persistent	19	4.75 (0.91)	4.31 (0.93)
Recovered	35	3.74 (1.05)	5.29 (0.64)
Control	37	3.67 (1.23)	4.90 (0.93)