

Research Article

A Lag in Speech Motor Coordination During Sentence Production Is Associated With Stuttering Persistence in Young Children

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Purpose: The purpose of this study was to determine if indices of speech motor coordination during the production of sentences varying in sentence length and syntactic complexity were associated with stuttering persistence versus recovery in 5- to 7-year-old children.

Methods: We compared children with persistent stuttering (CWS-Per) with children who had recovered (CWS-Rec), and children who do not stutter (CWNS). A kinematic measure of articulatory coordination, lip aperture variability (LAVar), and overall movement duration were computed for perceptually fluent sentence productions varying in length and syntactic complexity.

Results: CWS-Per exhibited higher LAVar across sentence types compared to CWS-Rec and CWNS. For the participants

who successfully completed the experimental paradigm, the demands of increasing sentence length and syntactic complexity did not appear to disproportionately affect the speech motor coordination of CWS-Per compared to their recovered and fluent peers. However, a subset of CWS-Per failed to produce the required number of accurate utterances.

Conclusions: These findings support our hypothesis that the speech motor coordination of school-age CWS-Per, on average, is less refined and less mature compared to CWS-Rec and CWNS. Childhood recovery from stuttering is characterized, in part, by overcoming an earlier occurring maturational lag in speech motor development.

Stuttering is a multifactorial neurodevelopmental disorder characterized by the presence of involuntary stuttering-like disfluencies (SLDs) during speech production, which are the overt result of disruptions in coordination patterns of neural commands to the muscle systems involved in speech (Ludlow & Loucks, 2003; Smith, 1989; Walsh & Smith, 2013). Stuttering has long been described as a disorder of speech motor discoordination and instability, with intervals of fluency and disfluency not observed as dichotomous phenomena, but instead as events along a continuum of speech motor coordination (Adams & Runyan, 1981; Van Riper, 1982; Zimmermann, Smith, & Hanley, 1981). Even when perceptually fluent, the speech of individuals who stutter has been associated with atypical speech motor coordination (Caruso, Abbs, & Gracco, 1988; McClean, 2004; McClean, Kroll, & Loftus, 1990; Zimmermann, 1980) and increased articulatory variability

(Kleinow & Smith, 2000; MacPherson & Smith, 2013; Smith, Goffman, Sasisekaran, & Weber-Fox, 2012) compared to typically fluent controls. Adults who stutter (AWS) have exhibited atypical speech and nonspeech motor performance compared to their typical peers, specifically in coordination tasks between different motor components or effector systems (Forster & Webster, 2001; Zelaznik, Smith, Franz, & Ho, 1997). Stuttering has also been associated with limitations in motor learning (Bauerly & De Nil, 2011; Smits-Bandstra & De Nil, 2007, 2009), and in performance of a nonword repetition task, AWS showed immature speech motor learning patterns (Smith, Sadagopan, Walsh, & Weber-Fox, 2010).

Although recent evidence from a study of bimanual clapping performance from our laboratory does not support a generalized motor disorder in young children who stutter (CWS; Hilger, Zelaznik, & Smith, 2015), previous studies have revealed atypical speech motor control processes in CWS close to onset (e.g., Smith et al., 2012; Walsh, Mettel, & Smith, 2015). Walsh et al. (2015) reported that in short, simple sentence production (e.g., “buy bobby a puppy”), relative to their typically fluent 4- and 5-year-old peers, CWS lagged in speech motor coordination and in measures of basic kinematic parameters, such as articulatory speed. In

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addition, in an experimental paradigm identical to the one employed in the current study with sentences of varying length and complexity, 4- to 6-year-old CWS exhibited increased lip aperture variability (LAVar) compared to children who do not stutter (CWNS) for fluent productions (MacPherson & Smith, 2013). Such results suggest that CWS in the preschool years lag behind their peers in speech motor development. Critical questions that follow from such findings are whether this early lag in speech motor coordination is associated with the chronicity of stuttering and what effect increasing linguistic demands, such as sentence length and syntactic complexity, have on the speech motor control processes of children who persist in, versus recover from, stuttering.

Maturational Indices of Speech Motor Development

Fluent speech is produced by numerous dynamic interactions between articulatory, laryngeal, and respiratory subsystems along with the rapid formation of articulatory patterns generated from neural commands to muscles (Levelt, 1989). Speech motor development is characterized by increasingly stable and mature speech patterns fostered by the emergence of functional synergies, or coordinated muscle collectives, which incorporate different motoric systems (Smith, 2006). Coordinated patterns of articulatory movement that are consistent form and stabilize during a protracted development toward increasingly efficient and robust speech motor control that decreases in variability into the teenage years and beyond (Dromey, Boyce, & Channell, 2014; Smith & Zelaznik, 2004; Walsh & Smith, 2002).

The development of one functional synergy, lip aperture, represents the movement coordination of the upper lip, lower lip, and jaw necessary for speech. Smith and Zelaznik (2004) examined the development of lip aperture during sentence production in individuals from age 4 years to early adulthood. The authors found that the dynamic control of lip aperture, the distance between upper and lower lips, stabilizes and becomes less variable during sentence production as children mature. However, this maturational progression is not linear; motor variability fluctuates at different points in development as a function of the rate of increasing motoric ability (Green, Moore, Higashikawa, & Steeve, 2000; Green, Moore, & Reilly, 2002; Smith & Zelaznik, 2004). These findings are consistent with dynamic systems and motor programming views of motor development, which propose that decreasing variability is a maturational trend towards increasing stability in the coordination of motoric subsystems (Thelen & Smith, 1994) through the organization of functional synergies of motor units at the periphery and the stabilization of feedforward-based motor programming circuits centrally (Guenther & Vladusich, 2012).

One useful method of quantifying this dynamic development of speech motor control processes is the computation of a spatiotemporal index (STI; Smith, Goffman, Zelaznik, Ying, & McGillem, 1995). The STI measures the consistency of interarticulator coordination over repeated

utterances (Smith, Johnson, McGillem, & Goffman, 2000). A consistent pattern of similarly repeated time- and amplitude-normalized productions denotes low variability in speech movement patterning, a characteristic of a highly practiced and stable speech motor system seen in typical adults (Maner, Smith, & Grayson, 2000). High STI scores indicate an unstable speech system that is less efficient in motor planning and execution (Smith & Goffman, 1998) and undergoing a period of learning and development (Goffman, 2010). Decreasing variability in articulation has been associated with advances in language development (Grigos, 2009). The linguistic demands of increasing sentence length and syntactic complexity conversely have a destabilizing effect on speech motor control in both typical children (Maner et al., 2000) and adults (Kleinow & Smith, 2006).

These findings suggest a relationship between language and speech motor processes, with articulatory variability either increasing or decreasing according to the linguistic competency of the child and linguistic complexity of the speech task. Previous findings from our laboratory suggest that both indices of language processing (e.g., Usler & Weber-Fox, 2015) and speech motor coordination (e.g., MacPherson & Smith, 2013) are atypical and immature in CWS compared to typically developing children. The demands of utterance length and syntactic complexity have also been correlated with the likelihood and loci of SLDs during speech production (Logan & Conture, 1995; Yaruss, 1999; Zackheim & Conture, 2003).

Purpose of the Study

The purpose of our study was to (a) determine if indices of speech motor coordination and movement duration during perceptually fluent speech are associated with the persistence of stuttering versus recovery from stuttering in 5- to 7-year-old children and (b) determine if the increasing linguistic demands of sentence length and syntactic complexity have a disproportionate effect on the speech motor systems of children with persistent stuttering (CWS-Per) compared to children who have previously recovered (CWS-Rec) and CWNS. We predicted that CWS-Per would likely exhibit higher LAVar, but not longer movement duration, compared to CWS-Rec and CWNS, on the basis of findings from previous studies (MacPherson & Smith, 2013; Walsh et al., 2015). We also hypothesized that increasing linguistic complexity would have a disproportionate effect on the speech motor systems of CWS-Per compared to our other two groups.

Method

Participants

With parental consent, our participants were enrolled in the Institutional Review Board (IRB)-approved Purdue Stuttering Project; many had taken part in previous studies from our laboratory (e.g., MacPherson & Smith, 2013;

Usler & Weber-Fox, 2015). For this greater project, children were followed longitudinally over a maximum of 5 years, during which time annual testing was conducted in as close to 12-month intervals as possible. All participants underwent initial evaluations at 4–5 years of age. Data for this current study, from 73 native English-speaking children between the ages of 5;9 (years;months) and 8;0, were collected during either the participants' first, second, or third annual visits to our laboratory. Six of our 73 subjects failed to successfully complete our sentence production task and were excluded from this study; five of these six children were CWS-Per and one child was from the CWNS group. On the basis of stuttering diagnosis and history, children ($N = 67$) were assigned to one of three groups: children whose stuttering was persisting at time of testing ($n = 21$; 17 boys, four girls; CWS-Per), children who had previously recovered from stuttering ($n = 15$; 10 boys, five girls; CWS-Rec), and children who do not stutter ($n = 31$; 20 boys, 11 girls; CWNS). Mean age (in months) was similar across groups: CWS-Per ($M = 78.19$, $SE = 1.31$), CWS-Rec ($M = 78.40$, $SE = 1.15$), and CWNS ($M = 77.61$, $SE = .73$). Mean age of stuttering onset, determined by parent report, was 34.47 months ($SE = 2.48$) for CWS-Rec and 35.33 months ($SE = 2.03$) for CWS-Per.

The presence of stuttering was determined on the basis of criteria established by Yairi and Ambrose (2005). After initial recruitment and classification, participants underwent subsequent annual evaluations to determine if they met the criteria for either stuttering persistence or recovery. Recovery from stuttering required that three criteria were all met at time of testing: (a) the child was identified as fluent by a parent and project clinician; (b) stuttering severity was rated under 2 on an 8-point severity scale by a project clinician or parent; and (c) the child produced fewer than three SLDs per hundred syllables during a spontaneous speech sample consisting of 750–1,000 words over two sessions (parent/child and clinician/child). SLDs were quantified using Yairi and Ambrose's (2005) weighted stuttering index, with the presence of dysrhythmic phonations (such as prolongations and blocks) having greater weight compared to repetitions during computation of stuttering severity. None of the CWS-Rec participants, to our knowledge, exhibited a return of stuttering behaviors in future years.

Participants did not exhibit any neurological deficits or symptoms of autism according to the Childhood Autism Rating Scale (Schopler, Reichler, & Renner, 1986). A history of medication for attention-deficit/hyperactivity disorder was reported by two CWS-Per participants at the time of testing. All participants passed a hearing screening at 20 dB HL for 500, 1000, 2000, 4000, and 6000 Hz. Socioeconomic status (SES) for each participant was assessed on the basis of the level of maternal education on Hollingshead's education scale (Hollingshead, 1975). Using this scale, medians for CWS-Rec (range = 4–7) and CWS-Per (range = 4–7) were rated at 6, which is equivalent to completion of a 4-year university education. The median rating for CWNS was 7, with a range from 5–7. A non-parametric independent-samples median test revealed group

differences in SES ($p = .01$). CWNS did not differ significantly in SES with CWS-Rec ($p = .53$), but did with CWS-Per ($p = .004$). There was no significant difference in SES between CWS-Rec and CWS-Per ($p = .17$).

All participants demonstrated normal nonverbal intelligence as assessed by the Columbia Mental Maturity Scale (CMMS; Burgemeister, Blum, & Lorge, 1972), with no significant differences between groups, $F(2, 64) = 2.85$, $p = .07$. Language comprehension was tested using the Test for Auditory Comprehension of Language—Third Edition (TACL-3; Carrow-Woolfolk, 1999). The Consonant Inventory subtest of the Bankson-Bernthal Test of Phonology (BBTOP-CI; Bankson & Bernthal, 1990) was used to assess phonological ability. Expressive language ability was tested using the Structured Photographic Expressive Language Test—Third Edition (SPELT-3; Dawson, Stout, & Eyer, 2003). All of our participants scored within the normal range on this battery of assessments, except for one CWS-Rec and two CWS-Per participants who scored below normal limits on the BBTOP-CI. Scores were significantly different across groups on the SPELT-3, $F(2, 64) = 5.63$, $p = .01$; and the TACL-3, $F(2, 64) = 3.77$, $p = .03$. Post hoc Bonferroni analyses revealed that CWS-Rec did not differ from CWS-Per or CWNS in the SPELT-3 ($p > .05$), but CWS-Per differed significantly from CWNS ($p = .01$). CWS-Rec similarly did not differ in TACL-3 performance from CWNS ($p = .26$) and CWS-Per ($p = 1$), but CWS-Per and CWNS did differ ($p = .03$). Group differences in performance on the BBTOP-CI were not statistically significant, $F(2, 64) = 1.63$, $p = .20$. See Table 1 for participant characteristics for the CWS-Per group. Group means and statistical comparisons for CMMS, TACL-3, SPELT-3, and BBTOP-CI are presented in Table 2.

Stimuli and Procedure

Participants were told they would hear sentences about birds and butterflies through a speaker, and to repeat the sentences exactly. A picture of birds and butterflies was displayed on a computer monitor located in front of the participant, concurrently with the presentation of a model production of the sentence. Four declarative sentences containing a large number of bilabial consonants (so that lip aperture is a dynamically controlled articulatory target) and varying in length and syntactic complexity were presented. As shown in Table 3, short sentences contained six words (eight syllables) and long sentences contained nine words (11 syllables). Sentences that are syntactically complex included a late-developing subject relative clause, previously shown to influence variability in speech motor coordination (Kleinow & Smith, 2006). The long sentences differed from their short counterparts through the addition of a nonobligatory adjunct.

During initial practice trials, production was modeled and feedback was given. Following this, stimuli were presented in blocks consisting of eight quasi-randomized sentences, resulting in participants producing six to 10 error-free and fluent productions of each sentence. Following

Table 1. Children with persistent stuttering (CWS-Per) participant characteristics

| Participant | Sex | Age at testing (in months) | Age of onset (in months) | Family history | Stuttering behaviors | Weighted stuttering index | Stuttering severity | Therapy exposure |
|-------------|-----|----------------------------|--------------------------|----------------|----------------------|---------------------------|---------------------|------------------|
| CWS-Per 1 | M | 73 | 30 | Y | SS, PW, DP, T | 1.94 | 2 | S, A |
| CWS-Per 2 | M | 76 | 48 | N | SS, PW, DP | 18.41 | 5.5 | S, A |
| CWS-Per 3 | M | 74 | 36 | N | SS, PW, DP, T | 3.12 | 2.5 | None |
| CWS-Per 4 | M | 73 | 36 | Y | SS, PW, DP, T | 4.07 | 2 | None |
| CWS-Per 5 | M | 83 | 36 | Y | SS, PW, DP, T | 4.46 | 3.5 | S |
| CWS-Per 6 | M | 74 | 24 | N | SS, PW, DP | 3.13 | 2 | None |
| CWS-Per 7 | M | 87 | 48 | N | SS, PW | 1.65 | 3 | None |
| CWS-Per 8 | M | 83 | 36 | N | SS, PW, DP | 6.56 | 2.5 | S |
| CWS-Per 9 | M | 77 | 54 | N | SS, PW, DP | 8.29 | 5 | S |
| CWS-Per 10 | F | 74 | 30 | U | SS, PW, DP | 4.45 | 3 | S, A |
| CWS-Per 11 | M | 81 | 36 | N | SS, PW | 1.32 | 2 | None |
| CWS-Per 12 | M | 78 | 24 | N | SS, PW, DP | 2.11 | 1 | A |
| CWS-Per 13 | M | 80 | 30 | N | SS, DP | 3.98 | 3 | None |
| CWS-Per 14 | M | 82 | 24 | Y | SS, PW | 5.52 | 3.5 | S |
| CWS-Per 15 | F | 72 | 36 | U | SS, PW, DP | 1.04 | 1 | S |
| CWS-Per 16 | F | 77 | 24 | Y | SS, PW, DP, T | 9.55 | 3 | S |
| CWS-Per 17 | M | 79 | 52 | Y | SS, PW, DP, T | 25.69 | 6(p) | S |
| CWS-Per 18 | M | 80 | 36 | N | SS, PW, DP | 8.93 | 6 | S |
| CWS-Per 19 | F | 96 | 36 | N | SS, PW, DP, T | 3.38 | 2 | S |
| CWS-Per 20 | M | 73 | 42 | N | SS, DP | 1.42 | 3 | S, A |
| CWS-Per 21 | M | 70 | 24 | N | SS, DP | 6.17 | 5 | None |

Note. Age of onset (in months) reported by parent. Family history = Yes (Y), No (N), Unknown (U). Stuttering behaviors = single syllable whole word (SS), part-word repetition (PW), dysrhythmic phonation (DP), tension (T). Weighted stuttering index = weighted number of stuttering-like disfluencies/100 syllables (see Yairi & Ambrose, 2005) at time of testing. Stuttering severity = stuttering severity rating (0–7) provided by clinician or parent (p) at time of testing. Therapy exposure before or during time of testing for stuttering (S), articulation (A). M = male; F = female.

the child's production of a sentence, a 2- to 3-s pause occurred before the next stimulus. The stimuli and data collection procedures of the current study were identical to those used in a previous study by MacPherson and Smith (2013).

Data Measurement and Analysis

During the repeated sentence productions, superior-inferior movements of the upper lip and lower lip were recorded using a Northern Digital Optotrak 3020 system (Northern Digital [NDI], Waterloo, Ontario, Canada). Previous studies from our laboratory have used similar

measurement and analysis methods (e.g., Kleinow & Smith, 2006; Smith & Zelaznik, 2004; Walsh, Smith, & Weber-Fox, 2006). Kinematic measures included LAVar and movement duration (in seconds). The Optotrak system tracked the motion of eight infrared light emitting diodes (IREDs), each sampled at 250 samples per second, in three dimensions, with an accuracy of 0.1 mm. IREDs were attached to the midline vermilion borders of each participant's upper and lower lips to track lip motion. Participants also wore a modified sports goggle with four attached IREDs. A fifth IRED was placed on the center of the forehead. These five markers were used to create a 3D coordinate system to track overall head motion. Head movement artifact

Table 2. Group mean and standard errors for nonverbal IQ and language assessment scores.

| Test | CWS-Per | CWS-Rec | CWNS | Group statistics |
|----------|---------------|---------------|---------------|------------------------------|
| | M (SE) | M (SE) | M (SE) | |
| CMMS | 112.57 (2.32) | 106.53 (1.78) | 113.71 (1.84) | $F(2, 64) = 2.85, p = .07$ |
| TACL-3 | 114.14 (1.74) | 116.27 (3.15) | 122.42 (2.25) | $F(2, 64) = 3.77, p = .03^*$ |
| SPELT-3 | 105.33 (2.26) | 107.20 (2.87) | 113.67 (1.36) | $F(2, 64) = 5.63, p = .01^*$ |
| BBTOP-CI | 97.90 (1.53) | 99.87 (2.57) | 102.00 (1.39) | $F(2, 64) = 1.63, p = .20$ |

Note. CWS-Per = children with persistent stuttering; CWS-Rec = children who had recovered from stuttering; CWNS = children who do not stutter; CMMS = Columbia Mental Maturity Scale; TACL-3 = Test for Auditory Comprehension of Language—Third Edition; SPELT-3 = Structured Phonological Expressive Language Test—Third Edition; BBTOP-CI = Banks-on-Bernthal Test of Phonology—Consonant Inventory.

*Statistically significant ($p \leq .05$; one-way ANOVA test).

Table 3. Sentence stimuli.

| Sentence length | Syntactic complexity | Sentence | Number of words, syllables | Relative clause |
|-----------------|----------------------|--|----------------------------|-----------------|
| Short | Simple | The birds and the butterflies played. | 6, 8 | Absent |
| Short | Complex | The birds that saw butterflies played. | 6, 8 | Present |
| Long | Simple | The birds and the butterflies played by the pond. | 9, 11 | Absent |
| Long | Complex | The birds that saw butterflies played by the pond. | 9, 11 | Present |

was removed from superior–inferior lip motions. Using a condenser microphone, the Optotrak system also recorded the acoustic signal from the repeated productions. The audio signal was digitized at 16,000 samples per second after low-pass filtering with a cutoff frequency of 7500 Hz. Significant differences in LAVar and movement duration were considered using an alpha level of $p \leq .05$. Repeated measures analyses of variance with Bonferroni post hoc corrections were used to determine main and interaction effects. Partial eta squared (η_p^2) was used to determine the size of significant effects.

Measurement of Lip Aperture Variability

Articulatory movements were measured using a custom, interactive MATLAB program (MathWorks, Natick, MA) that displayed lip displacement and velocity signals for each production. The recording of upper and lower lips began with the release of the /b/ in “birds” in all sentences and ended with the release of the /p/ in “played” for the short sentences and in “pond” for the long sentences. Upper and lower lip displacement signals, verified with the acoustic signal, were segmented into analysis records from the points of peak velocity of these first and last bilabial movements of each sentence production.

Lip aperture signal reflects the coordinated activity of muscles of both lips and jaw to control this important vocal tract parameter as a function of time. Each set of six to 10 lip aperture signals for each production was time- and amplitude-normalized. As described in earlier studies, time normalization was achieved by linearly interpolating all records to 1,000 points (e.g., Smith et al., 1995). Amplitude normalization was achieved by subtracting the mean and dividing by the standard deviation of each record. These standard deviations were calculated at 2% intervals in relative time and summed to create a LAVar index. The LAVar index is a quantitative measurement of the degree of variability in the coordination between the upper lip and lower lip across multiple productions. A higher LAVar index indicates inconsistent articulatory patterns over multiple productions. Figure 1 illustrates the steps in kinematic data processing, including the displacement signals (top panel), time-normalized signals (middle panel), and LAVar indices (bottom panel) for a single participant from each group.

Measurement of Movement Duration

Movement duration was computed as the time (in seconds) of each original, nonnormalized sentence (see Figure 1, top panel), starting at the release of the /b/ in “birds” and ending at the release of the /p/ in “played” for the short

sentences or the /p/ in “pond” for the long sentences. The durations of repeated productions for each sentence were averaged for every participant before statistical comparisons were performed.

Results

Lip Aperture Variability

A condition effect was observed for Sentence Length, with higher LAVar for longer sentences, $F(1, 64) = 50.45$, $p < .001$, $\eta_p^2 = .44$. A condition effect was also observed for Syntactic Complexity, with higher LAVar for complex sentences, $F(1, 64) = 11.46$, $p = .001$, $\eta_p^2 = .15$. A main effect of Group was observed, $F(2, 64) = 6.84$, $p = .002$, $\eta_p^2 = .18$. LAVar across the sentence types was similar between CWS-Rec and CWNS ($p = 1$). To the contrary, CWS-Per exhibited higher LAVar compared to CWNS ($p = .002$) and CWS-Rec (a trending significance of $p = .08$). We did not observe any Group interactions with Sentence Length, $F(2, 64) < 1$, or Syntactic Complexity, $F(2, 64) < 1$. Also, no Group \times Sentence Length \times Syntactic Complexity effect was observed, $F(2, 64) = 1.38$, $p = .26$. See Figure 2 for mean LAVar indices for CWS-Per, CWS-Rec, and CWNS across sentence types. Individual differences in LAVar were observed in all sentence types. Considerable overlap in LAVar between CWS-Per, CWS-Rec, and CWNS participants for the production of long, complex sentences is revealed in Figure 3.

As reported in the Method section, a nonparametric sample median test revealed a statistical difference in SES across our groups. We also observed significant group differences in performance on the SPELT-3 and TACL-3. However, there were no significant differences in SES, SPELT-3, and TACL-3 between CWS-Rec and CWS-Per. When these scores were individually included as covariates in our ANOVA analyses, no significant interactions with LAVar were observed ($p > .05$). In addition, the performances of all participants on the SPELT-3 and TACL-3 were within the normal range.

Movement Duration

A condition effect was observed for Sentence Length, $F(1, 64) = 2,202.55$, $p < .001$, $\eta_p^2 = .97$, and Syntactic Complexity, $F(1, 64) = 336.51$, $p < .001$, $\eta_p^2 = .84$. Movement duration for short sentences was predictably shorter ($M = 1.69$ s, $SE = 0.02$ s) compared to long sentences ($M = 2.40$ s, $SE = 0.03$ s). Duration was also shorter for syntactically

Figure 1. Kinematic data processing steps. Original lip aperture (LA) signals (top panel), normalized lip aperture signals (middle panel), and lip aperture variability indices (bottom panel) for short, complex sentence productions by one child who does not stutter (CWNS; left column), one child who has recovered from stuttering (CWS-Rec; center column), and one child with persistent stuttering (CWS-Per; right column). Different colors distinguish individual trials.

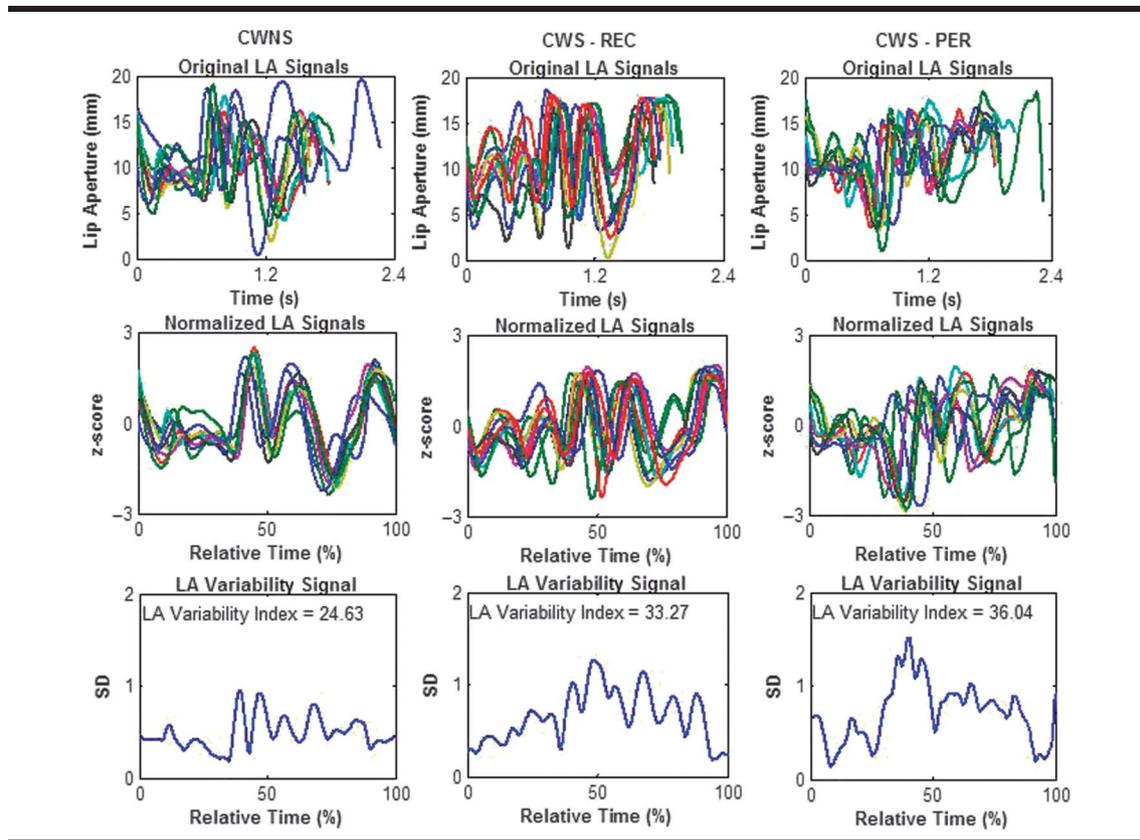


Figure 2. Lip aperture variability (LAVar) indices for children with persistent stuttering (CWS-Per), children who have recovered from stuttering (CWS-Rec), and children who do not stutter (CWNS) across all sentence types. Symbols represent means. Error bars indicate standard error.

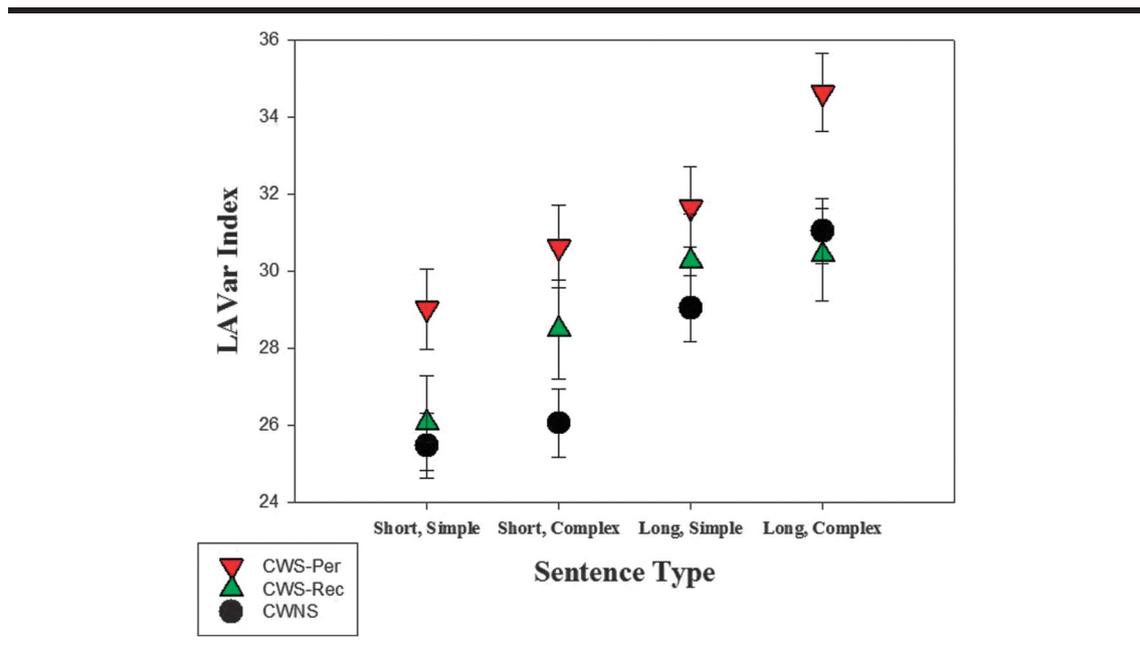
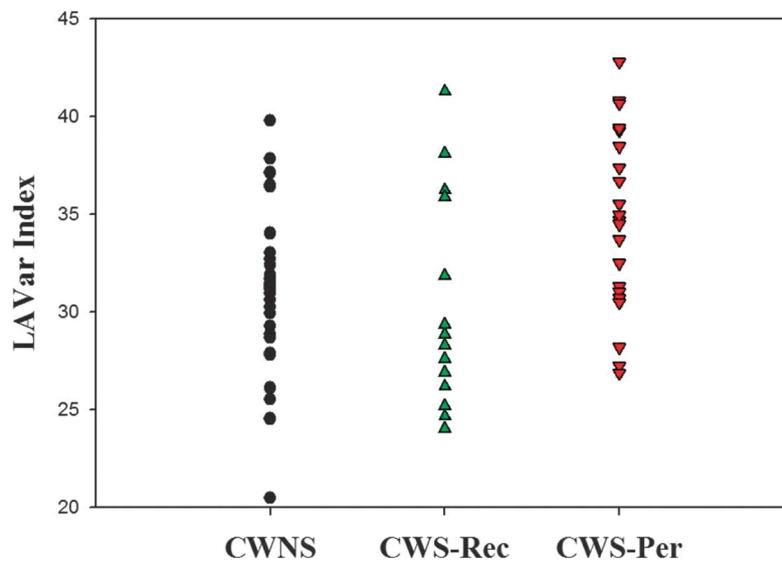


Figure 3. Individual children with persistent stuttering (CWS-Per), children who have recovered from stuttering (CWS-Rec), and children who do not stutter (CWNS) data for production of long, complex sentences. Please note that, in some cases, the number of visible data points per group may be less than the actual number due to the overlap of multiple symbols. LAVar = lip aperture variability.



simple sentences ($M = 1.89$ s, $SE = 0.03$ s) compared to syntactically complex sentences ($M = 2.20$ s, $SE = 0.03$ s). Movement duration did not differ among groups for any of the sentence types: A Group effect was not observed, $F(2, 64) = .78, p = .47$. As with LAVar, we observed neither a Group \times Sentence Length, $F(2, 64) < 1$, nor a Group \times Syntactic Complexity interaction, $F(2, 64) = 2.03, p = .14$. No Group \times Sentence Length \times Syntactic Complexity interaction was observed, $F(2, 64) = 1.78, p = .18$. Movement duration was also not significantly correlated with LAVar for any group across sentence types ($p > .05$; CWS-Per r range = $-.11$ to $.39$, CWS-Rec r range = $-.32$ to $.39$, CWNS r range = $-.10$ to $.19$). See Table 4 for group mean movement duration times across the sentence types.

Relationship Between Kinematic Indices and Stuttering Severity

Pearson correlational analyses were performed to determine any associations between our kinematic indices

and stuttering severity. A severity rating was given to each child by a project clinician using an 8-point scale. A weighted stuttering index was also determined on the basis of the number of SLDs per hundred syllables of spontaneous speech. A medium association between stuttering severity rating and the LAVar of long, complex sentences was observed (Pearson correlation, $r = .35, p = .04$). All other correlations were not significant ($p > .05$).

Discussion

Persistence of Stuttering at 5–7 Years of Age Is Associated With a maturational Lag in Speech Motor Coordination Ability

We examined the speech motor coordination of 5- to 7-year-old CWS-Per, CWS-Rec, and CWNS during the repeated production of sentences varying in length and syntactic complexity. LAVar distinguished CWS-Per from CWS-Rec and CWNS across the sentence types. On the

Table 4. Group mean movement duration times (in seconds, with standard errors) across sentence types.

| Group | Short simple sentence | Short complex sentence | Long simple sentence | Long complex sentence |
|---------|------------------------|------------------------|------------------------|------------------------|
| | <i>M</i> (<i>SE</i>) |
| CWS-Per | 1.49 (.04) | 1.80 (.04) | 2.25 (.05) | 2.49 (.06) |
| CWS-Rec | 1.54 (.03) | 1.87 (.04) | 2.24 (.04) | 2.55 (.05) |
| CWNS | 1.53 (.03) | 1.89 (.04) | 2.27 (.05) | 2.63 (.06) |

Note. CWS-Per = children with persistent stuttering; CWS-Rec = children who had recovered from stuttering; CWNS = children who do not stutter.

other hand, movement duration was comparable in our three groups. These results are not due to group differences in SES or language abilities. Our findings correspond with the MacPherson and Smith (2013) results of higher LAVar, but not movement duration, in CWS compared to CWNS. CWS-Per, on average, tend to exhibit higher LAVar compared to the recovered and fluent children, and similar LAVar was observed for these latter two groups. Although the present study was not longitudinal, higher LAVar is an indicator of a possible maturational lag in speech motor development (e.g., Smith & Zelaznik, 2004). To the contrary, maturation of speech motor coordination to a degree similar to that of typical children appears to be a characteristic of recovery from stuttering by 5–7 years of age.

The mechanism underlying the group differences in LAVar is likely the atypical temporal and spatial coding of the neural commands to muscles necessary for fluent speech production (Smith, 1989) and not orofacial characteristics such as bone mass or muscle physiology (Walsh & Smith, 2013). Possible sources for the higher movement variability of CWS-Per include variability in motor planning (Churchland, Afshar, & Shenoy, 2006) and execution (Smith, 1989), aberrant neural synchrony and deficits in the use of sensory feedback (Kurz, Heinrichs-Graham, Arpin, Becker, & Wilson, 2013), and the amount of neuromotor noise during movement execution (Harris & Wolpert, 1998). Neuromotor noise arises from brain areas responsible for motor planning and execution, and may fluctuate due to movement complexity, cognitive-linguistic load, and other stressors (Churchland et al., 2006; Van Beers, Haggard, & Wolpert, 2004; Van Gemmert & Van Galen, 1997). The central nervous system of CWS-Per, on average, may exhibit increased levels of neuromotor noise compared to CWS-Rec and CWNS, resulting in difficulty using sensory information to optimally execute a motor plan (Wolpert, 2007). Stuttering has been associated with limitations in sensorimotor integration (Hickok, Houde, & Rong, 2011; Max, 2004; Max, Guenther, Gracco, Ghosh, & Wallace, 2004; Namasivayam & van Lieshout, 2011), including a deficit in the use of feed-forward processing during speech (Cai et al., 2012; Civism, Tasko, & Guenther, 2010).

Effect of Sentence Length and Syntactic Complexity on the Speech Production of CWS-Per

Contrary to our hypothesis that increasing linguistic demands would have a disproportionate effect on the speech motor systems of CWS-Per, the effects of sentence length and syntactic complexity on LAVar and movement duration were parallel across the groups. The speech motor stability of CWS-Per who were able to complete our speech motor task (completing ≥ 6 accurate and fluent sentence productions), compared to their recovered and fluent peers, was not more susceptible to breakdown in the face of increasing linguistic demands. This finding was surprising given previous evidence that articulatory coordination variability of adults and typically developing children is affected by utterance length and syntactic complexity

(Kleinow & Smith, 2000; Smith et al., 2012). The lack of group differences may be because our analysis, similar to that of MacPherson and Smith (2013), only included CWS with relatively robust speech motor coordination. Not included in our analysis were five CWS-Per who failed to accurately produce the required number of long, complex sentence productions.

Theoretical Implications

Our findings support the view that developmental stuttering is a heterogeneous, neurodevelopmental, and multifactorial disorder involving atypical speech motor control processes (Smith, 1999; Smith & Kelly, 1997). Despite considerable individual differences, variability in articulatory coordination distinguished children who had recovered from those who persist in stuttering. Although coordination variability increased comparably for all groups with increasing linguistic demands, CWS-Per exhibited an unstable speech motor system (as indexed by LAVar) compared to their recovered and fluent peers.

If speech motor development involves the maturation of increasingly efficient and robust coordination patterns (Smith & Zelaznik, 2004), the higher variability in CWS-Per may represent an immature, flexible speech motor system. Neuromotor patterning to oral muscles of school-age CWS-Per remain unstable, evidence that these children are in a dynamic state of speech motor development. Stable neuromotor patterns facilitating fluent speech, represented by lower LAVar, conversely have developed in CWS-Rec and CWNS. Such a developmental trajectory in children who are persisting in stuttering is consistent with the ultimate characteristics of persistent stuttering in adults. They show immature patterns of lip aperture coordination in a nonword learning task (Smith et al., 2010) and when producing utterances of increased length and syntactic complexity (Kleinow & Smith, 2000). The conclusion that some 5- to 7-year-old CWS-Per have a developmental lag in speech motor control parallels previous findings with preschool-age CWS (MacPherson & Smith, 2013; Walsh et al., 2015). Our findings also support recent neuroimaging studies reporting atypical gray matter development in Broca's area in people who stutter (Beal et al., 2015) and developmental differences between CWS and CWNS in white matter connectivity in the inferior frontal gyrus (Chang, Zhu, Choo, & Angstadt, 2015).

Still, natural recovery during childhood is likely not driven solely by a maturation of speech motor processes. Bidirectional interactions between language formulation and speech motor coordination ability have been observed (Dromey & Bates, 2005; Goffman, 2010; Smith & Goffman, 2004). Recent studies from the Purdue Stuttering Project have reported maturational lags in various aspects of language processing related to stuttering persistence versus recovery (Kreidler, Hampton Wray, Usler, & Weber, 2016; Mohan & Weber, 2015; Usler & Weber-Fox, 2015). Under a dynamic view of development, the unstable speech motor control of some CWS-Per may be an adaptive effect

of a maturational lag in neurodevelopment underlying language acquisition. Although speculative, the relatively greater speech motor plasticity of some CWS-Per may indicate a period of dynamic speech-language learning and movement exploration (Goffman, 2010; Wu, Miyamoto, Castro, Ölveczky, & Smith, 2014). In contrast, the speech motor abilities of CWS-Rec have stabilized to the level of typically developing fluent children.

Study Limitations

This study did not control for a number of variables that may have an effect on the speech motor behavior of the participants, including a lack of control over the time between stuttering onset and testing, and the time since recovery for CWS-Rec. Many of the CWS-Per and CWS-Rec participants also reported participation in speech therapy for stuttering and/or other language deficits.

Conclusion

The purpose of this study was to determine if speech motor coordination processes were associated with stuttering persistence versus recovery in 5- to 7-year-old children. Articulatory coordination variability increased comparably for all groups with sentence length and syntactic complexity. There was no difference in coordination between children who have recovered from stuttering and typically developing children. Yet, CWS-Per exhibited higher overall coordination variability compared to the other two groups, even for production of a short, simple sentence. A subset of CWS-Per were also more prone to fail at accurately and fluently producing the required number of long, complex sentences to complete the task. The higher articulatory coordination variability in CWS-Per reveals that, unlike their recovered peers, these children at 5–7 years of age exhibit a maturational lag in their speech motor development, which evidence from previous studies from our laboratory suggests started in the preschool years (Walsh et al., 2015). However, we observed considerable individual performance differences in all three groups, with each child going through an individualized developmental landscape of speech motor coordination improvement. Still, stuttering from the beginning is often associated with a lag in speech motor development, and recovery is associated with “catching up” to their typically developing peers.

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