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Language and motor abilities of preschool children who stutter: Evidence from behavioral and kinematic indices of nonword repetition performance

Anne Smith[a,*], **Lisa Goffman**[a], **Jayanthi Sasisekaran**[b], and **Christine Weber-Fox**[a]

[a]Speech, Language, and Hearing Sciences, Purdue University, United States

[b]Speech-Language-Hearing Sciences, U. Minnesota, United States

Abstract

Stuttering is a disorder of speech production that typically arises in the preschool years, and many accounts of its onset and development implicate language and motor processes as critical underlying factors. There have, however, been very few studies of speech motor control processes in preschool children who stutter. Hearing novel nonwords and reproducing them engages multiple neural networks, including those involved in phonological analysis and storage and speech motor programming and execution. We used this task to explore speech motor and language abilities of 31 children aged 4–5 years who were diagnosed as stuttering. We also used sensitive and specific standardized tests of speech and language abilities to determine which of the children who stutter had concomitant language and/or phonological disorders. Approximately half of our sample of stuttering children had language and/or phonological disorders. As previous investigations would suggest, the stuttering children with concomitant language or speech sound disorders produced significantly more errors on the nonword repetition task compared to typically developing children. In contrast, the children who were diagnosed as stuttering, but who had normal speech sound and language abilities, performed the nonword repetition task with equal accuracy compared to their normally fluent peers. Analyses of interarticulator motions during accurate and fluent productions of the nonwords revealed that the children who stutter (without concomitant disorders) showed higher variability in oral motor coordination indices. These results provide new evidence that preschool children diagnosed as stuttering lag their typically developing peers in maturation of speech motor control processes.

Educational objectives—The reader will be able to: (a) discuss why performance on nonword repetition tasks has been investigated in children who stutter; (b) discuss why children who stutter in the current study had a higher incidence of concomitant language deficits compared to several other studies; (c) describe how performance differed on a nonword repetition test between children who stutter who do and do not have concomitant speech or language deficits; (d) make a general statement about speech motor control for nonword production in children who stutter compared to controls.

Keywords

Stuttering; Pre-school children who stutter; Nonword repetition task; Speech motor processes; Language and motor interactions

*Corresponding author. Tel.: +1 765 494 7743., asmith@purdue.edu (A. Smith)..

1. Introduction

The typical course of development of language production skills occurs over an extended period of time from infancy through the teenage years and entails learning at many different levels: auditory perceptual, linguistic, and sensorimotor. In a variety of developmental disorders, speech and/or language development follows an atypical course, and over the last two decades, the nonword repetition (NWR) task has proved to be a valuable experimental tool for exploring the nature of developmental speech and language disorders (Deevy, Wisman Weil, Leonard, & Goffman, 2010; Gathercole, 2006). A primary focus of this research has been on the nonword repetition skills of children with specific language impairment (SLI, Bishop, North, & Donlan, 1995; Dollaghan & Campbell, 1998; Gathercole & Baddeley, 1990), and poor performance of children with SLI, especially as nonword length increases, is a hallmark of this disorder (see Graf Estes, Evans, & Else-Quest, 2007, for a recent review). These results initially were interpreted to suggest that children with SLI have poor phonological storage capacities, because nonword repetition engages short-term phonological working memory (Gathercole & Baddeley, 1989, 1990). Recent accounts, however, have emphasized that performance on nonword repetition tasks also depends upon auditory processing, phonological analysis, and speech motor planning and execution, in addition to phonological storage (Gathercole, 2006; Shriberg et al., 2009). Interestingly, children with SLI lag their normally developing peers in speech and general motor skills (Bishop & Edmundson, 1987; Goffman, 1999; Zelaznik & Goffman, 2010) and SLI often has been associated with poorer auditory processing skills (e.g., Benasich & Tallal, 2002). Thus the weak performance of children with SLI on nonword repetition may reflect multiple aspects of the underlying disorder.

Stuttering is a developmental speech disorder in which the primary symptoms are motor; the flow of fluent speech is disrupted as the nervous system fails to generate the appropriate command signals to drive the muscles involved in speech production. Many accounts of the onset and development of stuttering, including our multifactorial, dynamic account (Smith, 1999; Smith & Kelly, 1997), posit that multiple factors interact to produce the breakdowns in speech production that we perceive as disfluencies (Conture et al., 2006; Van Riper, 1971; Wall & Myers, 1995). In these accounts, linguistic, emotional, and motor processes are proposed to interact in a manner that affects the probability of occurrence of speech motor breakdowns, and these factors in turn are determined by the interaction of one’s genetic makeup and experience. On the other hand, there are also theoretical accounts of stuttering in which the motor problems are viewed as “downstream” effects resulting from a core linguistic processing deficit (Perkins, Kent, & Curlee, 1991; Postma & Kolk, 1993). The covert repair hypothesis of Postma and Kolk specifically pinpoints slow and erroneous phonological encoding as the root cause of stuttering. Clearly, the NWR task would provide a useful window onto the processing levels potentially affected in children who are stuttering. Yet, there have been few earlier studies of nonword repetition skills in children who stutter (CWS).

Hakim and Ratner (2004) reported a preliminary study in which they compared eight CWS within a large age range (4;3 to 8;4 years) to age-matched fluent control participants using the Children’s Test of Nonword Repetition (CNrep; Gathercole, Willis, Baddeley, & Emslie, 1994). CWS had fewer correct productions and more phonemic errors than normally fluent speakers for one-, two-, and three-syllable nonwords, but significant group differences were observed only at the three-syllable level. A higher percent of phoneme errors was observed in both groups for the longer, four- and five-syllable nonwords. Anderson, Wagovich, and Hall (2006) compared performance of 12 CWS and age-matched controls between 3 and 5 years of age on the CNrep (Gathercole et al., 1994). CWS exhibited significantly fewer correct productions of 2- and 3-syllable nonwords and a higher percent of phonemic errors in the three-syllable nonwords of the CNrep compared to the typically developing children. The authors of these two studies concluded that CWS have weaker phonological working memory skills compared to typically developing children. Anderson et al. (2006) also discussed the many levels of processing involved in the NWR task and noted that the weaker performance of the children who stutter may reflect atypical auditory discrimination skills and/or poorer speech motor planning and execution abilities.

A study of nonword repetition in 12 Iranian CWS aged 5–7 years and 12 controls (Bakhtiar, Ali, & Sadegh, 2007) was designed to test the covert repair hypothesis (Kolk & Postma, 1997; Postma & Kolk, 1993). Bakhtiar et al. (2007) reported that the mean number of phonemic errors was not significantly different between the groups. They also compared reaction times between the two groups and found no difference between the stuttering and nonstuttering children, thus finding no support for the basic tenets, slowed and/or erroneous phonological encoding, of the covert repair hypothesis. In a study of ERP responses in a rhyme judgment task in older, school-age children (Weber-Fox, Spruill, Spencer, & Smith, 2008), we included a NWR task. We also found no differences in performance on the NWR task (Dollaghan & Campbell, 1998) between the groups of ten CWS and matched, normally fluent children aged 9–13 years.

From this review it is clear that nonword repetition abilities of CWS are not well understood, and the available findings are mixed. It is also important to note that CWS often show concomitant language and/or phonological disorders (Arndt & Healey, 2001; Blood, Ridenour, Qualls, & Hammer, 2003; Conture, 1990; Louko, Edwards, & Conture, 1990) or reduced language abilities without frank deficits (Ntourou, Conture, & Lipsey, 2011), but this has been a matter of some debate (Nippold, 2001). Contributing to the lack of clarity on this issue is the fact that various investigators have used different tests to evaluate the language status of their participants, and in most cases these tests have not probed morphosyntactic abilities (e.g., the PPVT-III; Dunn & Dunn, 1997 is a test of receptive vocabulary). In any case it is clear that language status would strongly affect the nonword repetition skills of CWS. Therefore, a major objective of the present experiment was to assess nonword repetition skills in CWS whose speech and language abilities have been carefully documented. In addition to assessing the children's behavioral performance on a NWR task (Dollaghan & Campbell, 1998), another goal of this research was to directly examine speech motor output during nonword production. This is an important level of analysis to include, because speech motor processes are obviously essential components of the nonword production process.

There have been few earlier studies in which the effects of linguistic demands on speech motor output measures (e.g., articulatory movement or muscle activity) have been examined in CWS. The speech motor systems of *adults* who stutter (and indeed their motor systems in general) show greater signs of instability compared to normal speakers, even when their speech is perceptibly fluent, and they show greater instability in the face of increased length and syntactic complexity of utterances (e.g., Kleinow & Smith, 2000; McClean, Kroll, & Loftus, 1990; Zimmermann, 1980).

In a recent study of adults who stutter, we (Smith, Sadagopan, Walsh, & Weber-Fox, 2010) employed a NWR task to further probe the speech motor dynamics of adults who stutter. The nonwords varied in length and phonological complexity of the segmental constituents (e.g., "mab," "mabshibe," "mabshaytiedoib"). The nonwords were presented auditorily in random order, so that each participant produced each nonword approximately ten times over the course of the experimental session. We also administered a standardized nonword repetition test (Dollaghan & Campbell, 1998) to the participants. There were no differences between the groups in the accuracy of their nonword repetition. The design of this experiment allowed us to examine potential within-session practice effects, that is, decreasing movement variability and increasing movement speed with practice (Schmidt & Wrisberg, 2004). Practice effects are potentially important to consider, because they reveal the underlying dynamics of speech motor learning. With regard to assessing speech motor processes, the NWR task is ideal, because presumably participants are generating novel speech motor programs to produce phonetic sequences for the novel nonwords. Adults who stutter showed practice effects in measures of coordination and speed of production. Their interarticulatory coordination variability decreased in the later compared to the earlier productions, and their nonword production durations also decreased. The normally fluent adults, on the other hand, showed no improvement in the consistency of interarticulatory coordination over the session, presumably because their initial productions were consistently well coordinated. The normally fluent adults did show practice effects evidenced by increased speed of nonword production.

The behavioral findings from this study of nonword repetition in adults who stutter generally do not support theories of stuttering that propose a phonological processing deficit as the primary causal factor in stuttering (Kolk & Postma, 1997). The adults who stutter could listen to novel nonwords, retain them in phonological working memory, and generate and execute a motor plan to produce the novel sequences fluently. The NWR task proved sensitive to differences in the speech motor dynamics underlying the fluent speech of adults who stutter. In the motor output, clear differences were seen between the adults who stutter and the control participants. The speech motor variability of the adults who stutter was significantly higher than that of the normally fluent adults, and it was strongly affected by the length and phonological complexity of the nonword. Adults who stutter showed practice effects within the experimental session that were not characteristic of the normally fluent speakers; rather the response of the adults who stutter to the task was similar to that observed in an earlier study using the same protocol in typically developing 9- and 10-year-olds (Walsh, Smith, & Weber-Fox, 2006). In general the results of this experiment support the notion that, compared to normally fluent speakers, adults who stutter have vulnerable speech motor systems as indicated by greater instability in the face of increased processing demands.

In the present study, we employ the same nonword repetition protocol used in our earlier study of adults who stutter to examine speech motor dynamics in preschool CWS and matched participants who do not stutter. Our goal was to expand on the results of earlier behavioral studies of nonword repetition abilities in preschool CWS and their typically developing peers by using specific and sensitive measures of speech and language abilities to classify subgroups of CWS with and without concomitant speech sound and/or language impairments. In addition, we used kinematic measures to elucidate the potential effects of increasing utterance length and complexity on the speech motor performance of these young children.

2. Methods

2.1. Participants

Thirty-one preschool CWS (24 males) and 22 children who did not stutter (CWNS, 12 males) participated in the study. Table 1 provides characteristics of the two groups of children. The data were collected as part of a large-scale, longitudinal study of language and speech motor skills in CWS and their CWNS peers. Data collection was carried out at two sites, the Department of Speech, Language, Hearing Sciences, Purdue University and the Department of Communication Sciences and Disorders, University of Iowa. All participants were native speakers of North American English. All participants passed a hearing screening at 20 dB HL at 500, 1000, 2000, 4000, and 6000 Hz and had normal or corrected-to-normal vision as per parent report. All children were free of neurological disorders and had no history of taking medications that may affect cognitive function (for example, medications for depression, seizures, or attention-deficit hyperactivity disorder). They also demonstrated normal non-verbal intelligence as assessed by the Columbia Mental Maturity Scale (Burgemeister, Blum, & Lorge, 1972); the range and median of these scores were similar for the stuttering and normal fluent groups (see Table 1). The children showed no symptoms of impaired reciprocal social interaction and restriction of activities (DSM IV criteria of autism and pervasive developmental disorder: American Psychiatric Association, 1994) as assessed by the Childhood Autism Rating Scale (Schopler, Reichler, & Renner, 1988), and based on parental report had no prior history of treatment for emotional problems.

We recorded the level of mothers' education as an index of the child's socioeconomic status (Hollingshead, 1975). On this scale a score of 3 indicates that the mother did not complete high school, 4 – graduated high school, 5 – partial college or specialized training, 6 – completed college degree, and 7 indicates a graduate degree. As indicated in Table 1, the two groups were well matched on this index of socioeconomic status.

2.2. Stuttering diagnosis

Participants were classified as CWS if all of the following three criteria developed by Ambrose and Yairi (1999) were met: (1) the child was regarded as having a stuttering problem by an experienced speech-language pathologist working on this project; (2) the child's stuttering severity was rated as 2 or higher on an eight point severity scale by either his/her parents or a speech-language pathologist; (3) the child exhibited at least three stuttering-like disfluencies (SLDs) per 100 syllables of spontaneous speech. Spontaneous speech samples were analyzed from a parent–child interaction and from a clinician–child interaction. Research assistants were trained by a speech pathologist with many years of experience in child language disorders and stuttering to code the types (stuttering-like or normal) and numbers of disfluencies in the spontaneous speech samples and reached a minimum reliability of 85% between 2 trained listeners.

2.3. Speech and language testing

Speech production skills were tested using the Bankson–Bernthal Test of Phonology (BBTOP; Bankson & Bernthal, 1990). Six children in the stuttering group scored below the normal range (<85) on the consonant inventory (CI) scale of the BBTOP. Grammatical aspects of language production were assessed using the Structured Photographic Expressive Language Test – Edition 3 (SPELT-3; Dawson, Stout, & Eyer, 2003). This test is a sensitive and specific measure of SLI (Perona, Plante, & Vance, 2005) that taps morphosyntactic skills. Nine of the 31 children who stutter scored below expected levels (standard score < 85) on the SPELT. Receptive vocabulary and syntax were evaluated using the Test for Auditory Comprehension of Language – Edition 3 (TACL-3; Carrow-Woolfolk, 1999). All of the children who stutter passed the TACL. The Appendix provides characteristics of the 31 CWS including the results of the speech and language testing which were used to form subgroups of CWS with and without concomitant speech sound and/or language disorders.

Experimental results are reported for three subgroups of stuttering children: children who stutter without concomitant language or speech sound disorder (CWS), children who stutter who have a concomitant speech sound but not language disorder (CWS+SS), and children who stutter with a concomitant language disorder (CWS+LI). The CWS+LI category includes some children who also have speech sound deficits; speech deficits often co-occur in children with SLI (Deevy et al., 2010). Means (and SDs) of language and speech performance for each group are presented in Table 2. Independent sample t-tests were computed to compare each group of stuttering children with their CWNS peers. There were no significant differences between the CWS and CWNS groups on the three language tests. As indicated in Table 2, the CWS+SS differed from the CWNS group only on the BBTOP, $t(26) = 8.8$, $p < .001$. The CWS+LI scored lower than the CWNS group on all three speech and language tests (BBTOP, $t(29) = 5.6$, $p < .001$; SPELT, $t(29) = 8.5$, $p < .01$; TACL, $t(29) = 2.2$, $p < .04$).

2.4. Other testing

2.4.1. Working memory skills—Verbal and nonverbal working memory measures were obtained using the auditory digit and word span tests of the Test of Auditory Perceptual Skills – Revised (TAPS-R; Gardner, 1996) and the nonverbal color, and order subtests (Color Block Non-Verbal Memory Task, Goffman, 2002, Purdue University, West Lafayette, IN). Compared to the CWNS group, the three groups of stuttering children tended to have lower mean scores on these tests of working memory, but the differences were significant only for the CWS+LI vs. CWNS comparison on the auditory number forward, $t(27) = 2.0$, $p = .05$, and backward, $t(27) = 2.4$, $p = .02$, tests.

2.4.2. Standard transcription measure of nonword repetition performance—Nonword repetition skills were assessed using the Nonword Repetition Test (NRT, Dollaghan & Campbell, 1998). Consistent with the standard administration procedure, the nonwords were recorded by a native English speaker and were presented over a speaker. Children were told that they would hear “silly” words and were asked to repeat each nonword. Scores are computed as percent phonemes correct in the one-syllable, two-syllable, three-syllable, and four-syllable nonwords. Phonemes were scored as correct whether produced fluently or disfluently, so that stuttering status did not affect the scores. The tests were scored by a graduate student in Speech-Language Pathology. During the training of the graduate student, the nonword repetition tests also were scored by a senior speech pathologist with over 25 years experience testing young children’s speech and language abilities. Training continued until the graduate student achieved 85% or higher agreement with the senior clinician.

2.5. Kinematic data collection

2.5.1. Kinematic measure of nonword repetition performance—Length was manipulated in a set of four nonwords – “mab” (/*mæb*/, 1 – syllable), “mabshibe” (/*mæbʃaIb*/, 2 – syllables), “mabfieshabe” (/*mæbfaiʃeIb*/, 3 – syllables), “mabshaytiedoib” (/*mæbʃeItaIdɔIb*/, 4 – syllables). The nonwords differed in the number of constituent syllables (1–4) and in the number and complexity of the phonemic elements to create increased phonemic complexity. An additional nonword, “mabteebeebee” (/*mæbtibibi*/), consisting of four syllables but with a predominance of bilabial consonants that are reduplicated and acquired early during speech development, was employed to assess potential differences in motor execution when nonword length, but not complexity was manipulated. The first consonant of the first syllable/*mæb*/and the final consonant (/*b*/) were identical for all nonwords. This strategy was used to enable selection of consistent start and end points for articulatory movement data extraction on the basis of lower lip peak opening velocities during the articulatory trajectory analysis (Smith, Johnson, McGillem, & Goffman, 2000). The stimuli, produced by an adult female who is a speaker of native American English, were recorded and digitized using PRAAT software (Boersma & Weenink, 2012). For all nonwords primary stress was on the first syllable.

2.5.2. Apparatus—Participants were seated in front of the Northern Digital Optotrak 3020 cameras, a commercial system that allows tracking of movements in 3D with an accuracy of <0.1 mm. Eight small (7 mm) infra-red light emitting diodes (IREDs) were attached to track articulatory movements of the upper lip, lower lip, and jaw. Four of the IREDs were mounted on a set of goggles that participants wore during the experimental session. One IRED was placed in the center of the forehead. Together, these five IREDs were used to calculate the 3D head coordinate system (Smith et al., 2000), which allowed head movement artifact to be eliminated. To track the motion of the lips, one IRED was placed at the center of the vermilion border of the upper lip and one at the center of the vermilion border of the lower lip (this marker represents combined actions of the lower lip and jaw). The IRED motions were sampled at a rate of 250 samples/s. A condenser microphone was used to record the speech signal, which was digitized on an A/D channel of the Optotrak system and was thus synchronized with the movement data. The acoustic signal was low-pass filtered with a cut-off frequency of 7500 Hz and digitized at 16,000 samples/s.

2.5.3. Kinematic experimental protocol—As in the Dollaghan and Campbell (1998) NWR task, participants were told that they would hear “funny, made-up” new words that they had not heard before. They were asked to listen carefully and repeat each novel word as best as they could. They were instructed to direct their gaze toward two soft toy dogs that were placed in front of them, on top of the Optotrak cameras used to track the articulatory movements. Following instructions, each nonword was presented via loudspeakers during practice trials. Any errors were corrected by the experimenter with a maximum of three practice trials. Following this, the child attempted to repeat each nonword without assistance for two correct trials. Following two correct productions, or if after three practice trials, two correct productions had not been produced, practice was terminated, and kinematic data collection began.

The experimental session lasted approximately 30 min. During the session, the nonwords were presented auditorily in 8 blocks of 10 nonwords. Each block consisted of the five nonwords presented twice in quasi-random order. Each trial was initiated with the experimenter playing a nonword, immediately followed by the child producing the target. This was followed by a 2–3 s pause before presenting the next nonword in the block.

2.6. Dependent variables

2.6.1. Behavioral analysis—Accuracy and fluency of the “mab” words was judged off-line by two observers. One, a graduate student in the clinical speech pathology program, coded the nonwords as fluent, a normal disfluency, ND, or a stuttering-like disfluency, SLD (Ambrose & Yairi, 1999). A second observer with over ten years of experience on the project performed the kinematic analysis. She listened to each nonword to decide if it was both fluent and accurate, requirements to be included in the kinematic analysis. The audio signal, which accompanied a computer display of the kinematic records, was played back for each token, and the experimenter decided if that token should be used in the kinematic analysis.

2.6.2. Kinematic measures—The lip aperture (LA) signal for each of the sets of correct and fluent nonword productions was computed as the distance between the lips as a function of time (upper lip signal minus lower lip signal in the superior-inferior dimension, which included jaw motion). Fig. 1 provides sample records of the lip aperture signal. This signal was used to compute two kinematic measures: movement duration and lip aperture variability. The overall movement duration associated with each nonword repetition was measured as the time between the initial and final oral opening movements. The LA variability index represents the degree of dispersion of the LA trajectories associated with multiple repetitions of each nonword. Accordingly, it is a measure of the degree of consistency in coordinating the upper lip, lower lip, and jaw (lower lip marker motion reflects contribution of both lower lip and jaw) over repeated productions of each nonword (Smith & Zelaznik, 2004). A higher value of the LA index indicates greater variability in coordinative patterns for repeated productions of each nonword. The methods for time and amplitude normalization of the lip aperture trajectories and for computing the lip aperture variability index are identical to those reported by Smith and Zelaznik (2004) and Walsh et al. (2006). Briefly, linear normalization allowed trajectories of the repetitions for each nonword to be plotted in normalized time (all records are linearly interpolated to 1000 points), and amplitude normalization was achieved by subtracting the mean and dividing by the SD for each LA trajectory. The LA variability index reflects the sum of 50 standard deviations computed at 2% intervals in relative time across the 10 normalized trajectories.

In order to reduce the amount of data lost, in some cases, eight or nine trials were used to compute the lip aperture variability index when 10 correct productions were not available. This was a reasonable strategy to adopt, because in unpublished work, we have observed a high correlation between kinematic variability indices based on 10 versus eight trials. Thus, for each nonword, the total number of children whose data were included in the analysis varied, depending on the number of children who were able to produce 8–10 correct repetitions. The LA variability index was based on eight or nine trials in 9.8% of 172 indices calculated, and these were approximately equally distributed among stuttering and normally fluent groups.

2.6.3. Practice effects—In order to examine potential practice effects, separate LA variability indices and movement durations were calculated for the first five trials and for the last five trials (using the 10 trials selected to calculate the overall indices above). Due to practice trials before kinematic data collection began and a variable number of total trials among the children depending upon the number of errors made, these subgroups of trials are referred to as “early” trials and “later” trials. In cases in which only 8 or 9 trials were used to compute the LA variability index, we used the first four and the last four as the early and later trials. If more than 10 fluent and accurate trials of the nonword were available, the first five trials and the last five trials were used (e.g., with 12 fluent and accurate productions, the early trials would be 1–5 and the later trials would be 8–12).

3. Results

3.1. Standard Nonword Repetition Test: effects of language status

In Fig. 2 percent phoneme correct scores for the four groups (CWNS, CWS, CWS+SS, and CWS+LI) on the Nonword Repetition Test (Dollaghan & Campbell, 1998) are plotted. From this graph it is clear that the performance of the CWS group was similar to CWNS, while the percent phoneme correct scores of the language impaired and speech sound disordered CWS were depressed, especially for the longer nonwords for the CWS+LI. Data for some nonword lengths, especially for the longer nonwords, were missing for 8 children. In order not to lose subjects, we elected not to do the planned repeated measures ANOVA. Instead, separate ANOVAs were computed to compare the groups for each nonword length (one-syllable through four-syllable). Table 3 provides the results of these ANOVAs and pairwise Fishers LSD post hocs for the three stuttering groups vs. the CWNS group. The CWS+SS group’s performance was significantly lower than the CWNS group only in 2-syllable nonwords; however the 1-syllable comparison narrowly missed significance. Predictably the CWS with concomitant language impairment showed the lowest percent correct phoneme scores overall, and the CWS+LI group performed with significantly lower accuracy for the one-syllable, two-syllable, and three-syllable nonwords. The four-syllable ANOVA did not reveal a significant group effect, as all the groups showed reduced accuracy on these nonwords, thus post hoc paired comparisons were not available for this nonword length.

3.2. Kinematic experiment results

3.2.1. Behavioral results for the "mab" word set—In order to be used in the kinematic data analysis for the "mab" word set, the production had to be accurate (i.e., no errors in entire word) and fluent. All groups of children were remarkably fluent during their production of the "mab" words. The median number of stuttering-like disfluencies and normal disfluencies was zero for all groups and all words, except in the case of the CWNS group on "mabshaytaidoib," the median number of normal disfluencies was 1.0. The maximum mean number of SLDs observed was 1.9 (average number of SLDs across children within a group for a single nonword), and this occurred for the CWS+LI on "mabshaytaidoib." The maximum mean number of normal disfluencies was 0.8 for the CWNS on "mabshaytaidoib." Therefore, in general, the fewer numbers of accurate and fluent trials available for the stuttering children for the kinematic analysis were not a result of large numbers of disfluencies; rather the excluded data arose from incorrect productions.

Table 4 lists the numbers of children in the CWNS, CWS, CWS+SS, and CWS+LI groups who produced at least 8 accurate, fluent productions of each nonword used in the kinematic analysis. The CWS with concomitant speech and/or language disorders did not produce accurate phonemic sequences beyond the one-syllable word, "mab." Therefore, insufficient data that met the criteria for the kinematic analysis were available for the CWS+SS and CWS+LI groups of children, and they were not included in the kinematic analysis. For the CWNS and CWS groups' performance was better, but only three CWS produced the requisite number of accurate trials of the most difficult nonword "mabshaytaidoib," so this word was dropped from the analysis. Finally the CWS who produced sufficient accurate data for the remaining nonwords were not always the same individuals (e.g., the seven who accurately produced at least 8 trials of "mabfaishabe" were not necessarily the same individuals who produced "mabteebeebee" correctly). Given the missing data for each word, we used a subset of data for the kinematic analysis (see below).

A relevant question is whether the small number of accurate nonword productions in the CWS+SS group was due to the fact that they typically omit or misarticulate phonemes contained in the nonwords. To address this question, we used the BBTOP results to assess the sounds that were not in the repertoires of each of the CWS+SS. Only one of the six children displayed an articulation error on a sound that was used in the "mab" nonword set. Thus we would conclude that the CWS+SS were capable of producing the sounds included in the nonword production task.

3.2.2. Lip aperture variability indices and trajectory duration—As noted above, numbers of correct and fluent trials varied among the participants with the CWS+LI and +SS groups having so few correct trials that they their kinematic data could not be analyzed. In order to employ the repeated measures ANOVA planned for comparisons between the CWS and CWNS groups, we used a subset of the kinematic data that maximized the number of subjects contributing data. For 13/22 CWNS and 8/16 CWS lip aperture variability indices were available for "mab," "mabshibe," and "mabteebeebee." These subsets of subjects and nonwords were included in the kinematic analysis. Mean LA variability indices obtained from the early and later trials for each of the three nonword are plotted in Fig. 3. A repeated measures ANOVA was computed with groups as the between factor and nonword and trials (early vs. later) as the within-subjects factors. CWS had higher LA variability indices, $F(1,19) = 4.63$, $p = .04$. As expected, the effect of word was significant, $F(2,38) = 58.9$, $p < .001$, but there was no word by group interaction. Although the plot in Fig. 3 suggests the presence of a practice effect (a reduction in lip aperture variability indices on later trials), especially for "mabteebeebee," the ANOVA was not significant for the effect of trials. There were no significant interactions of trial with group or word.

Noting that the two subgroups of CWS and CWNS included in this kinematic analysis differed in the percentage of males (CWNS, 50% male and CWS, 75% male) and that an earlier study of the same dependent variable showed that four and five-year old typically developing boys lag girls in speech motor development (Smith & Zelaznik, 2004), we wanted to determine if sex was a confound for the CWS vs. CWNS difference reported above. A repeated measures ANOVA revealed no effect of sex on the lip aperture variability indices for the three nonwords, $F(1,19) < 1$.

The total duration (ms) of the lip aperture trajectory of each nonword production was measured, and means of the early and later trials for the three nonwords are plotted for each group in Fig. 3. As the plots and Fig. 3 suggest, a repeated measures ANOVA revealed no differences between the CWS and CWNS groups, $F(1,19) < 1$, on trajectory durations, indicating that the two groups produced the nonwords at the same rate. The effect of word was significant, $F(2,36) = 300.7$, $p < .001$, but there was no word by group interaction. A practice effect was observed as a reduction in nonword duration on later trials, $F(1,19) = 10.9$, $p < .005$, but there were no significant interactions of trial with group, suggesting that both groups increased speech production rate in the later trials (Fig. 4).

4. Discussion

We used a NWR task to explore the interaction of language and motor abilities in preschool children who stutter and their normally fluent peers. The results of our experiment indicate that (1) children who stutter (CWS) who are not language impaired and do not have a speech sound disorder perform as well as their normally fluent peers on tests of nonword repetition; (2) Despite the behavioral accuracy of the CWS in performance of the NWR task, measures of oral motor coordinative variability for CWS suggest they lag their typically developing peers in speech motor development; (3) children who stutter who have a concomitant phonological disorder (CWS+SS) show poorer performance on nonword repetition tests compared to their CWNS peers and CWS with normal language skills; (3) children who stutter who have a concomitant language disorder (CWS+LI) show remarkably poor performance on nonword repetition tests compared to their CWNS peers and CWS with normal language skills, as has also been well documented in children with SLI who do not stutter (e.g., Bishop et al., 1995; Dollaghan & Campbell, 1998; Gathercole & Baddeley, 1990).

4.1. Behavioral results

Earlier studies of NWR skills in children who stutter produced equivocal results, with two studies finding no performance deficits in CWS aged 5–7 years (Bakhtiar et al., 2007) and in ten CWS aged 9–13 years (Weber-Fox et al., 2008) and two studies reporting significant differences between CWS and their CWNS peers in their production of two- and three-syllable nonwords (Anderson et al., 2006; Hakim & Ratner, 2004). On the surface, our results appear to be in conflict with those of Anderson et al. (2006), because they examined 12 children in the same age range as those assessed here, 3–5 year olds. They found that that children who stutter, who scored within expected levels on both the Peabody Picture Vocabulary Test – III (PPVT-III; Dunn & Dunn, 1997) and on phonological screening tests, produced fewer correct productions of two- and three-syllable nonwords. It is possible, we would suggest, Anderson et al. (2006) may in fact have failed to exclude stuttering children with concomitant SLI, because the PPVT taps receptive vocabulary skills. Many children with SLI perform well on receptive vocabulary measures, and it is morphosyntactic abilities that predominantly differentiate them from their normally developing peers (Leonard, 1998). We employed specific and sensitive measures of both grammatical language abilities (SPELT) and speech production abilities (BBTOP) in preschool CWS. Therefore the subgroup of CWS we identified that passed both of these tests were free of language deficits. However, they were stuttering, which reflects underlying deficits in speech motor processes. Thus, the present behavioral results suggest that accuracy on NWR tests is affected by language but not by fluency status.

One caveat should be mentioned relative to the behavioral findings. From the Dollaghan and Campbell (1998) NRT results, one would conclude that CWS and CWNS perform identically on all word length categories (Fig. 2). However, in the “mab” word set, we note that 10 of the 22 CWNS, or 45%, produced eight correct and fluent productions (the minimum number to perform the kinematic analysis) of the longest and most complex nonword, “mabshaytaidoib.” In contrast, only 3 of the 16 CWS, or 19%, produced this minimum number of correct productions. This result suggests that if linguistic and motor complexity increases, CWS without concomitant speech or language disorders may perform more poorly compared to their CWNS peers.

4.2. Kinematic results

The “mab” word set used in the kinematic investigation was designed to manipulate length and complexity of the phonetic elements of the nonwords. Sufficient data were available to perform the kinematic analysis for only 13 CWNS and 8 CWS for a subset of three of the five nonwords. CWS+SS and CWS+LI performed with very low accuracy on the “mab” nonword set; therefore these subgroups of CWS were excluded from any kinematic analysis. Only productions in which all elements are correct can be used in the lip aperture variability analysis, because combining trajectories underlying production of varying phonetic elements (e.g., “mabshibe” and “mabside”) could introduce potentially spurious variability. The analysis is designed to capture the variability in the underlying pattern of coordination when movement goals are constant (Smith et al., 2000). While it may be viewed as a weakness of our experimental design that the nonword set was so difficult for these young children, we created this specific stimulus set so that it would be challenging, and children would make errors. Also, we are following these children longitudinally, and as they get older and their overt speech errors diminish, we will be able to assess speech motor performance across all three groups.

From the subset of data we could analyze, we found that CWS have higher LA variability indices. This finding suggests that CWS are, on average, less consistent in speech motor planning and execution. We know of no other studies of oral motor control in young children who stutter. Thus, this is the first suggestion that CWS at age 4–5 years are lagging their CWNS peers in the consistency of coordination of their articulators. Both groups showed practice effects evidenced by an increase in rate of production of the nonwords in the later compared to the earlier trials. This is also a novel finding. Even during a short experimental session in which the children produce 12–15 tokens of each nonword (note that the five nonwords were randomly presented), the children increased their speech rate. Tested in precisely the same paradigm, CWNS children aged 9–10 years also sped up production of nonwords during the experiment, and they showed a practice effect by reducing coordinative variability in the later trials (Walsh et al., 2006).

In summary, we explored speech motor processes in preschool CWS, and we provide initial evidence of a lag in their speech motor development. These results, if confirmed in future studies in which more CWS complete the experimental task, suggest that the neural networks in the brain mediating speech motor processes are developing atypically and/or in a delayed manner in young children who are stuttering. It is interesting to note that approximately 50% of children who are stuttering at age four and five years will recover with or without therapeutic intervention (Watkins, Yairi, & Ambrose, 1999; Yairi & Ambrose, 1999). While the mean scores on oral motor coordination for nonword production were significantly higher in the CWS group in the current study, it is important to note that the scores of many of the CWS overlapped the normal range. A major goal of our longitudinal study is to determine if measures such as those presented here can be used to predict recovery versus persistence in young children who are stuttering. Finally, we acknowledge that our kinematic measure, which reflects the consistency of upper lip, lower lip, and jaw coordination, is only one of many that could be employed. Preliminary data from adults who stutter (Namasivayam & van Lieshout, 2008) suggest that other measures, for example of inter-gestural timing, may also reveal significant evidence of delays in speech motor development in children who stutter.

4.3. Implications for models of stuttering

Our findings indicate that four and five-year-old children who are stuttering are heterogeneous with regard to their language abilities, with some showing frank deficits and others scoring in the normal range on speech and language tests. Our results also suggest that children who are stuttering are more likely to be language impaired compared to the general population of preschool children; 7% of five-year-olds meet the diagnostic criteria for SLI (Tomblin et al., 1997). This finding should be interpreted with some caution, as our sample is not random, rather the parents self-selected, and the parents of CWS with concomitant speech and language disorders may be more likely to seek help. This higher incidence of concomitant speech and language disorders in stuttering children, however, is consistent with many earlier reports (Blood et al., 2003; Bloodstein & Ratner, 2008). We assessed 31 children diagnosed as stuttering. Only 50% of this sample scored in the normal range on all of the speech and language tests. Six of the 31, or 19% of the stuttering children in our sample had frank phonological deficits as indicated by their scores on the BBTOP, but they had normal language abilities. Nine CWS, 29% of our sample, failed the SPELT, indicating frank language impairment. These results support the assertion that the factors underlying stuttering in children have differing weights (Smith & Kelly, 1997; Van Riper, 1971). Clearly their phonological and/or expressive language deficits affected the CWS+SS and CWS+LI children’s performance on the NWR tasks. These subgroups produced so few correct productions of the nonwords used in the kinematic experiment that we could not explore the underlying motor dynamics. On the basis of earlier investigations (e.g., Munson, Edwards, & Beckman, 2005), we expected the children with phonological disorders to show reduced accuracy in NWR tasks; however, we were surprised that these children could not accurately produce even the relatively simple two- and three-syllable nonwords included in the nonword set designed to explore inter-articulator coordination. Poor performance on a test of nonword repetition is a hallmark of LI (Graf Estes et al., 2007), so our CWS+LI group’s inability to produce the longer and more complex nonwords was predictable.

We note above that approximately 50% of our sample of CWS showed frank phonological and/or language deficits. This is a similar proportion to that reported by Arndt and Healey (2001) on the basis of a survey of speech/language pathologists about their caseloads. The respondents indicated that 56% of their caseloads had a fluency disorder only and that 44% had verified concomitant phonological and/or language disorders, but the prevalence estimates are varied (Blood et al., 2003; Bloodstein & Ratner, 2008). The issue of co-occurrence of fluency and language or phonological disorders is also complicated by longitudinal studies demonstrating that children who are stuttering show varying language profiles at different sampling times, perhaps showing an early delay, then “catching up” (Paden, Ambrose, & Yairi, 2002; Paden, Yairi, & Ambrose, 1999). Again, such results are consistent with accounts of stuttering in which the various factors involved in producing stuttering have different weights in different individuals, and the weight of these underlying factors changes dynamically over the individual’s lifetime (Smith & Kelly, 1997; Van Riper, 1971).

The relatively high proportion of CWS identified as having frank phonological and/or language deficits in the present study is likely to be, at least partially, a result of the use of higher resolution measures of language abilities rather than omnibus screening tests. In future studies, it will be important to use more specific and sensitive measures, such as these, of phonological and language abilities (Plante & Vance, 1994). By carefully defining subgroups of stuttering children in the present experiment, we were able to determine that CWS who are clearly in the normal range in language/phonological abilities perform like CWNS children on tests of NWR. However, children who stutter who have concomitant phonological and/or language disorders do not perform as well as their CWS peers (without concomitant LI or SS disorders) and their CWNS peers.

With regard to models of stuttering, the present results provide strong evidence against theories of the etiology of stuttering that posit phonological encoding and/or retrieval as *the core* causal factor in the disorder, for example, the covert repair hypothesis (Postma & Kolk, 1993). However, CWS who do not have concomitant language or speech sound disorders can successfully engage all of the processing levels that are tapped in a NWR task, from auditory processing, to phonological analysis and storage, to speech motor planning and execution (Gathercole, 2006). Our results also support the hypothesis that phonological processing problems can play an important role in stuttering for some children. We also provide preliminary evidence that the output of the speech motor planning and execution processes are more variable in CWS compared to their CWNS peers. Further evidence inconsistent with models that target deficient and/or slowed phonological processing as the core cause of stuttering comes from the results reported by Bakhtiar and colleagues. They (Bakhtiar et al., 2007) found that the reaction times of CWS aged 5–7 years in a NWR task were equal to those of the CWNS controls. This finding suggests that CWS are not slower in performing the multiple processing stages engaged in a NWR task, albeit speech motor coordination for some of the CWS is more variable than that of their CWNS peers. Thus we conclude that, while phonological processing deficits may often be a contributing factor to the onset and development of stuttering, they are not the primary cause of stuttering in all children or adults who stutter.

Taking a different theoretical perspective, one may ask what the present findings tell us about the general abilities tapped by NWR tasks. The CWS without concomitant language or speech sound disorders performed with behavioral accuracy equal to their CWNS peers (although note the caveat above about the poorer performance of CWS in producing the longest and most complex nonword in the “mab” nonword set). Does this mean that speech motor planning and execution processes are not important in this task, given that these children who are stuttering and clearly have speech motor deficits, perform well? We would not draw this conclusion. Rather we would conclude from our findings and earlier studies (reviewed in Gathercole, 2006) that the NWR task relies heavily on language and phonological processing networks, and that the speech motor systems of our young CWS operated well enough, so that they could produce the nonwords. Both CWS and CWNS groups were challenged in similar ways by the length and complexity of the nonwords, and these challenges resulted in errors. In general, we would conclude from our results and earlier studies that stuttering, like SLI, is a complex, multidimensional, developmental disorder that potentially affects language processing and speech production at multiple levels.

CONTINUING EDUCATION

Language and motor abilities of preschool children who stutter: Evidence from behavioral and kinematic indices of nonword repetition performance

QUESTIONS

1. Nonword Repetition Tasks are hypothesized to engage all of the following processes EXCEPT:
   - **a.** Phonological storage
   - **b.** Semantic retrieval
   - **c.** Auditory processing
   - **d.** Speech motor planning
   - **e.** All of the above are engaged during a Nonword Repetition Task
2. The percentage of children who stutter (CWS) from the current study that had concomitant speech sound and/or language deficits was approximately:
   - **a.** 5%
   - **b.** 10%
   - **c.** 25%
   - **d.** 50%
   - **e.** 75%
3. Which of the following statements most accurately reflects how CWS performed on the Dollaghan and Campbell Nonword Repetition Test (1998):
   - **a.** CWS with speech sound deficits performed as well as CWS without speech/language deficits.
   - **b.** All CWS had significantly poorer scores compared to CWNS.
   - **c.** CWS without concomitant speech/language deficits performed as well as CWNS.
   - **d.** CWS with language impairment showed the poorest performance compared to CWNS.
   - **e.** Both C and D are correct.
4. Compared to the control participants, results from the study revealed that CWS (without concomitant speech or language deficits) had more variable:
   - **a.** Speech motor execution abilities
   - **b.** Phonological analysis abilities
   - **c.** Phonological storage abilities
   - **d.** Auditory processing
   - **e.** None of the above is correct
5. Kinematic analysis of nonword production revealed each of the following EXCEPT:
   - **a.** CWS had lower lip aperture variability indexes than CWNS.

b. A practice effect was observed as a reduction in nonword duration on later trials.

c. There was a significant effect of word on lip aperture variability indices.

d. There was no effect of sex on lip aperture variability indices.

e. CWS with concomitant speech/language deficits were not included in the analysis due to their poor performance.

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Appendix

Characteristics of the children who stutter. The label "CWS" indicates a child who stutters who passed the speech and language screening tests; "CWS+SS," a child who stutters with concomitant speech sound disorder; "CWS+LI," a child who stutters who has concomitant language and (in some cases) speech sound disorder.

Subject	Sex	Age (months)	SES	SLD[a]	BBTOP-CI	SPELT	TACL
CWS1	M	48	7	2.11	111	102	98
CWS2	M	49	6	6.29	110	105	91
CWS3	M	49	6	7.11	92	107	109
CWS4	F	58	7	1.95	102	110	124
CWS5	M	58	4	11.76	104	103	113
CWS6	F	59	4	4.41	115	122	102
CWS7	M	50	6	4.06	104	104	104
CWS8	F	51	7	2.73	108	102	117
CWS9	M	51	6	6.63	93	90	106
CWS10	M	52	4	6.1	113	95	109
CWS11	M	54	6	4.19	105	100	117
CWS12	M	55	6	2.47	100	111	96
CWS13	M	56	6	5.05	104	100	139
CWS14	M	57	6	1.68	99	102	111
CWS15	M	61	6	9.73	93	121	132
CWS16	M	66	7	2.84	108	113	115
CWS+SS17	M	48	6	10.34	76	115	132
CWS+SS18	M	58	6	7.05	72	116	143
CWS+SS19	M	50	6	5.4	72	87	117
CWS+SS20	M	60	6	4.8	77	102	130
CWS+SS21	M	61	5	2.85	81	93	117
CWS+SS22	M	67	4	7.82	72	92	121
CWS+LI23	F	49	4	14.46	86	85	96

Subject	Sex	Age (months)	SES	SLD[a]	BBTOP-CI	SPELT	TACL
CWS+LI24	F	49	6	4.93	88	77	106
CWS+LI25	M	48	7	4.78	76	68	102
CWS+LI26	M	48	4	2.25	82	82	109
CWS+LI27	F	49	4	19.97	85	82	126
CWS+LI28	M	59	4	9.34	93	80	104
CWS+LI29	F	49	6	5.69	85	82	113
CWS+LI30	M	55	6	8.09	69	84	100
CWS+LI31	M	72	6	3.47	103	84	115

[a]Someofthe CWS are recorded as having fewerthan three SLDs per 100 syllables, but they still met the criteria forthe stuttering diagnosis. This occurred, because the SLD measures listed are the result of averaging across the multiple 100 syllable segments of speech available from two conversational settings. If in one of these 100 syllable segments, the child exceeded three stuttering-like disfluencies, he or she was judged to meet this criterion forthe stuttering diagnosis (Yairi & Ambrose, 1999).

Biography

Anne Smith is a Distinguished Professor and has been investigating the physiological bases of stuttering for many years.

Lisa Goffman is a Professor in the department and an expert on language and motor interactions in typically developing children and children with SLI.

Jayanthi Sasisekaran is an assistant professor who studies language and speech motor interactions in stuttering.

Christine Weber-Fox is a Professor who studies the neural bases of language processing in children and adults.

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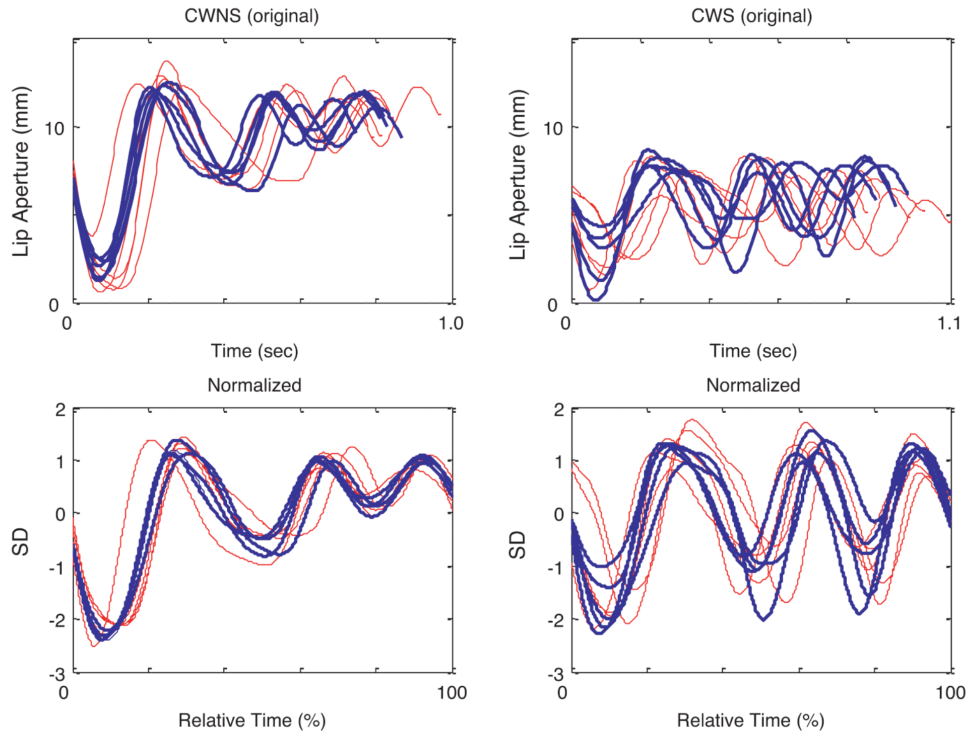

Fig. 1.
Sample kinematic data for "mabteebeebee" from a typically developing child, left column, and a child who stutters, right column. The top panel for each child shows the lip aperture trajectories for the sets of 5 early productions (thinner, red traces) and 5 later productions (thicker, blue traces). The top graphs show distance between the upper lip and lower lip (lip aperture signal) in mm as a function of real time. The lower plots show the same trajectories after time and amplitude normalization. From the two top plots, it is apparent that the two children are producing very different movement patterns. The CWNS child produced larger oral openings for the first syllable, whereas the CWS produced opening movements of about the same size for all four syllables. Also from the original data plots, we see that the red (early 5) trajectories tend to be of longer duration compared to the thicker, blue trajectories for both children, indicating the overall practice effect we observed: the later productions were produced faster. Also the blue trajectories tend to converge together in a tighter pattern compared to the early set. This suggests that the later productions were more consistently coordinated, and this should be reflected in lower lip aperture variability indices for the later compared to the early trials. This was the case. For the CWNS child the early trials LA Var was 18.7 and for the later trials 8.5; while for the stuttering child the indices were early 31.2 and later trials 21.4. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

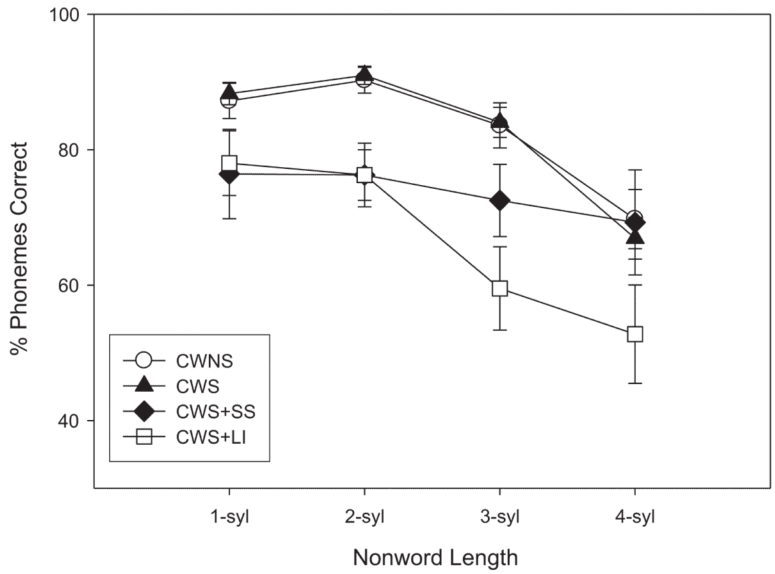

Fig. 2.

Percent phonemes correct (mean and SEM) in the Dollaghan and Campbell nonword repetition task (1998). CWNS, typically developing children; CWS, children who stutter who passed all language and speech screening test; CWS+SS, children who stutter with a phonological disorder; and CWS+LI, children who stutter with concomitant language disorder.

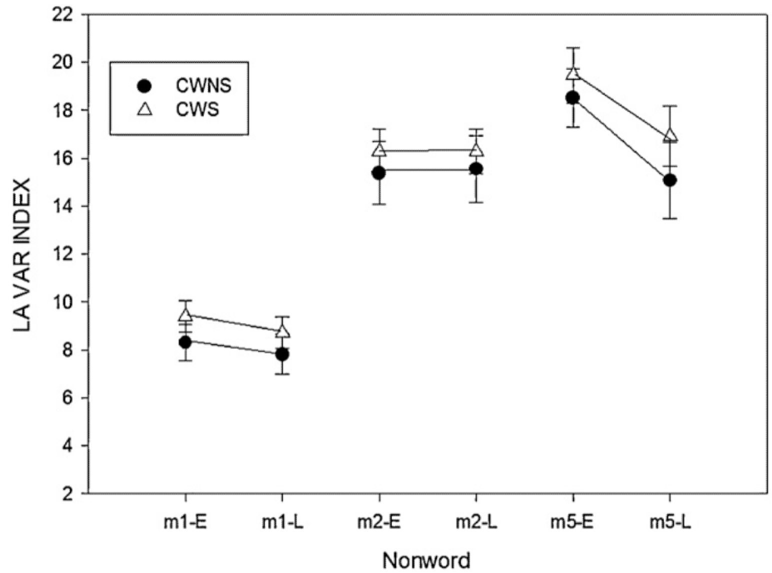

Fig. 3.
Lip aperture variability indices (mean and SEM) for the CWNS (typically developing children) and CWS (children who stutter) groups for m1, “mab,” m2, “mabshibe,” and m5, “mabteebeebee” for the early (E) and later sets of trials (L).

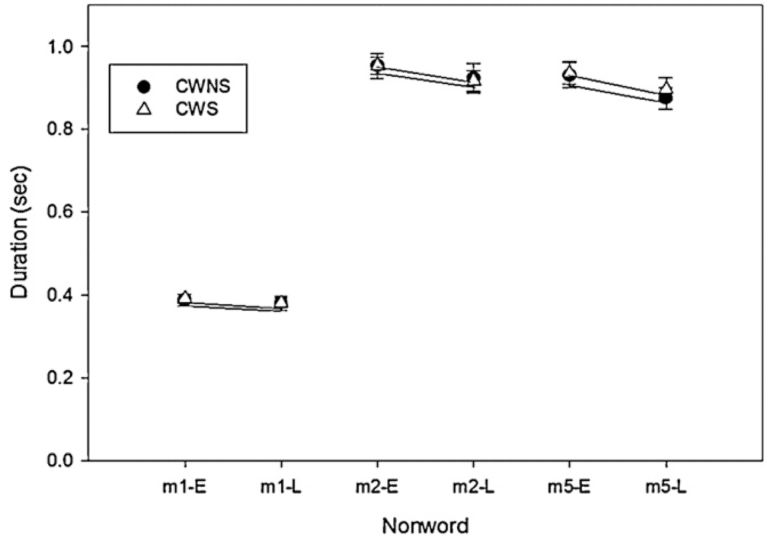

Fig. 4.
Overall duration (mean and SEM) for the CWNS (typically developing children) and CWS (children who stutter) groups for m1, “mab,” m2, “mabshibe,” and m5, “mabteebeebee” for the early (E) and later sets of trials (L).

Table 1

Characteristics of the stuttering and nonstuttering children.

	n (males)	Age (months)		SES		CMMS	
		Range	*Mdn*	Range	*Mdn*	Range	*Mdn*
CWNS	22(12)	48–71	59	3–7	6	91–127	110
CWS	31(24)	48–72	55	4–7	6	90–132	111

Table 2

Performance of children on speech and language tests. Children who are typically developing (CWNS), children who are stutter who passed the speech and language test (CWS), children who stutter with the concomitant phonological disorder (CWS+SS), and children who stutter with a concomitant language disorder (CWS+LI). Asterisks indicate significant differences between CWS+LI or CWS+SS and CWNS on each test. See text for detailed statistics.

	BBTOP-CI			SPELT			TACL		
	Range	**Mean**	**SD**	**Range**	**Mean**	**SD**	**Range**	**Mean**	**SD**
CWNS (22)	89–117	103	8.9	86–130	111	10.3	91–141	120	14.8
CWS (16)	92–115	104	7.1	90–122	105	8.4	91–139	111	12.8
CWS+SS (6)	72–81	75***	3.4	87–116	101	12.3	117–143	127	10.3
CWS+LI (9)	69–103	85***	9.6	68–85	80***	5.2	96–126	108*	12.0

* $p < .05$.

** $p < .01$.

*** $p < .001$.

Table 3

Results of four ANOVAs computed forthe percent phoneme correct scores (plotted in Fig. 2) for 1-syllable, 2-syllable, 3-syllable, and 4-syllable categories of the Dollaghan and Campbell (1998) NRT. The top row contains the *F*-value forthe between groups factor. The values in rows 2 through 4 are the Fishers' LSD probabilities (not available for 4-syllable because the main effect of group was ns) for paired comparisons.

	1-SYL	2-SYL	3-SYL	4-SYL
$F(3,45)$	3.18*	5.89**	5.80**	1.75
CWS/CWNS	$p = .58$	$p = .74$	$p = .90$	–
CWS+SS/CWNS	$p = .06$	$p = .01$**	$p = .11$	–
CWS+LI/CWNS	$p = .05$*	$p = .001$***	$p < .001$***	–

* $p < .05$.

** $p < .01$.

*** $p < .001$.

Table 4

Number of children in each participant group who produced at least 8 correct and fluent productions of each nonword in the kinematic experiment.

Group	mab	mabshibe	mabfieshabe	mabshaytiedoib	mabteebeebee
CWNS (*n* = 22)	21	14	12	10	14
CWS (*n* = 16)	15	10	7	3	8
CWS+SS (*n* = 6)	6	1	0	0	2
CWS+LI (*n* = 9)	8	2	1	0	2