

Research Article

Evidence That Bimanual Motor Timing Performance Is Not a Significant Factor in Developmental Stuttering

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Purpose: Stuttering involves a breakdown in the speech motor system. We address whether stuttering in its early stage is specific to the speech motor system or whether its impact is observable across motor systems.

Method: As an extension of Olander, Smith, and Zelaznik (2010), we measured bimanual motor timing performance in 115 children: 70 children who stutter (CWS) and 45 children who do not stutter (CWNS). The children repeated the clapping task yearly for up to 5 years. We used a synchronization-continuation rhythmic timing paradigm. Two analyses were completed: a cross-sectional analysis of data from the children in the initial year of the study (ages 4;0 [years;months] to 5;11) compared clapping performance between CWS and CWNS. A second, multiyear analysis assessed clapping behavior across the ages 3;5–9;5

to examine any potential relationship between clapping performance and eventual persistence or recovery of stuttering.

Results: Preschool CWS were not different from CWNS on rates of clapping or variability in interclap interval. In addition, no relationship was found between bimanual motor timing performance and eventual persistence in or recovery from stuttering. The disparity between the present findings for preschoolers and those of Olander et al. (2010) most likely arises from the smaller sample size used in the earlier study.

Conclusion: From the current findings, on the basis of data from relatively large samples of stuttering and nonstuttering children tested over multiple years, we conclude that a bimanual motor timing deficit is not a core feature of early developmental stuttering.

Stuttering is a speech production disorder characterized by disfluencies such as part-word repetitions, prolongations, and silent blocks. The etiology involves multiple factors, including motoric, linguistic, and psychosocial contributors (Conture, 1990; Smith, 1990; Starkweather, 1993; Van Riper, 1982; Wall & Myers, 1995). Stuttering onset generally occurs around 2–5 years of age with a 75% recovery rate for children who began to stutter in these years (Ambrose & Yairi, 1999; Watkins, Yairi, & Ambrose, 1999; Yairi & Ambrose, 1999). Stutter-like disfluencies result from disruptions in the neural commands to the muscles necessary for fluent speech (e.g., Smith, 1989). These motor disruptions are observed in articulatory, laryngeal, and respiratory systems (Kleinow & Smith, 2000; Max & Gracco, 2005; Max, Guenther, Gracco, Ghosh, & Wallace, 2004; McClean & Runyan, 2000; Peters & Boves, 1988; Smith, 1989;

Ward, 1997; Zimmermann, 1980). There is also evidence, though mixed, that children and adults who stutter (CWS and AWS, respectively) show less proficiency in nonspeech motor tasks (Brown, Zimmermann, Linville, & Hegmann, 1990; Caruso, Abbs, & Gracco, 1988; Cooper & Allen, 1977; Falk, Müller, & Bella, 2014; Forster & Webster, 2001; Howell, Au-Yeung, & Rustin, 1997; Max, Caruso, & Gracco, 2003; Neef et al., 2011; Olander, Smith, & Zelaznik, 2010; Ward, 1997; Webster, 1986; Westphal, 1933; Zelaznik, Smith, Franz, & Ho, 1997).

Extending the work of Olander et al. (2010), who found a large percentage of preschool CWS with atypical bimanual motor timing performance, we further investigate whether the neuromotor deficit involved in stuttering is reflected in general timing control. We assess nonspeech motor ability by measuring bimanual clapping performance in a larger sample of preschool children who were followed for 5 years in order to evaluate the potential interactions between manual motor timing control and eventual stuttering outcome. Based on the findings of Olander et al. (2010), we predict that a subset of CWS will show greater variability in nonspeech motor timing performance. From the

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longitudinal analyses of clapping performance, we examine maturation of clapping skill in groups of stuttering and nonstuttering children. Also, the longitudinal design allows us to determine which subgroups of CWS ultimately persisted or recovered. From this information, we can retrospectively classify preschool CWS as those who persisted (CWS_p) and those who recovered (CWS_r) to assess the prediction of Olander et al. (2010) that CWS who are poor in early general motor timing performance are more likely to persist in stuttering.

Atypical Nonspeech Motor Performance in Stuttering

It is hypothesized that some aspects of speech and nonspeech motor systems share underlying neural substrates; therefore, a deficit in one system (speech motor system) could be related to other systems (e.g., limb motor and/or nonspeech oral motor tasks). Measures of speech timing, vocal articulation, finger tapping, and foot tapping are positively correlated among similar movements of different effectors within individuals (i.e., speech articulators vs. finger movements), suggesting some degree of shared neural resources across motor systems (Cooper & Allen, 1977; Franz, Zelaznik, & Smith, 1992; Ivry & Richardson, 2002; Keele & Hawkins, 1982; Klapp, 1981). Therefore, it is possible that the speech motor deficit observed in stuttering may extend to nonspeech motor domains as well; however, as outlined below, previous studies of such tasks have produced mixed results.

Investigators who report a general timing deficit in stuttering have found that AWS demonstrate poor timing on a variety of paced and unpaced speaking, nonspeech oral motor tasks, and finger tapping tasks in which timing skills are measured by either the ability to synchronize with a beat, or the ability to accurately repeat a rhythm (Brown et al., 1990; Cooper & Allen, 1977; Forster & Webster, 2001; Ward, 1997). Chang, Kenney, Loucks, and Ludlow (2009) hypothesized that neural mechanisms for planning and execution of vocal tract gestures with auditory targets would be shared across speech and nonspeech movements in AWS and in controls. In an fMRI study, they assessed brain activation patterns in frontal premotor and temporoparietal regions and found that AWS showed different activation patterns both in the perceptual integration and planning and in the execution phases of speech and nonspeech tasks. The patterns of activation were different in the two groups, stuttering and control, but consistent within each group across speech and nonspeech tasks, supporting the hypothesized shared underlying neural control systems. In a recent transcranial magnetic stimulation study of the dorsolateral premotor cortex (PMd), subjects synchronized tapping the right index finger to a metronome (Neef et al., 2011). These investigators reported timing difficulties in 14 AWS after stimulation over the right PMd. In contrast, and as expected, AWNS exhibited timing difficulty with left PMd stimulation, suggesting atypical connectivity for timing of nonspeech movements in AWS. Evidence for a general coordination

deficit in stuttering also has been observed in AWS as greater variability in multimovement sequencing of lip, jaw, and finger movements using tasks such as accuracy in turning a crank, velocity of flexion movements, and accuracy of finger tapping sequences (Caruso et al., 1988; Forster & Webster, 2001; Max et al., 2003; Webster, 1986; Zelaznik et al., 1997). Thus a variety of investigations using measures of task performance as well as neuroimaging to examine neural activation patterns suggest that motor differences in AWS are not confined to speech production.

Some investigators, on the other hand, have failed to find differences in general timing and coordination processes in AWS. For example, Max and Yudman (2003) found that AWS showed similar timing abilities to the fluent controls across speech, orofacial nonspeech, and finger movements. They utilized the classic timing synchronization-continuation paradigm (Wing & Kristofferson, 1973a, 1973b) for repetitive movements in which 10 AWS and 10 AWNS matched their movements to a beat and then continued the rhythm in three tasks: speech, repeating the syllable /pa/ to measure bilabial movement; nonspeech oral, assessing oral opening and closing without the voiced speech component; and manual, successively moving the index finger to contact the thumb. Timing accuracy and variability did not differ between AWS and AWNS for all three tasks, suggesting that these AWS demonstrated normal timing ability. These findings are notable because multiple movement systems were examined. Last, two studies from our laboratory suggest that outcome differences may be affected by task complexity. Zelaznik, Smith, and Franz (1994) showed no simple timing deficits in AWS, but Zelaznik et al. (1997) found bimanual finger coordination deficits in the same group of AWS in a bimanual finger coordination task. In conclusion, in some cases AWS can demonstrate normal coordination and timing abilities across articulatory and manual motor systems, but in some cases their performance is atypical. The precise reasons for the inconsistencies in results of these studies are not clear, though they may be related to the heterogeneity of stuttering symptomology and the nature of the task under study.

Nonspeech Motor Ability in Childhood Stuttering

Examining motor abilities in children is a particularly important issue in relation to specifying potential factors that contribute to the onset and persistence of stuttering; however, few studies have examined nonspeech motor skills in CWS. In an early study, Westphal (1933) found that CWS 9 to 17 years of age scored lower than age-matched children who did not stutter (CWNS) on a number of motor tasks, including tossing beads and writing while blindfolded. Howell et al. (1997) reported that CWS (9–10 years) performed as well as CWNS on a sinusoidal lip-tracking task for timing accuracy but with greater variance. Falk et al. (2014) assessed motor timing in 20 CWS in two age groups, 10 children 8–11 years and 10 children 12–16 years, compared with 43 CWNS (22 younger children and 21 older children). They assessed the children's ability to synchronize

finger tapping to an external beat. Although the older CWNS demonstrated better timing performance than the younger CWNS, the older CWS did not perform better than the younger CWS, and overall, these children performed worse than their fluent age-matched peers. These findings suggest that the maturational course of improvement in motor timing control lags in CWS compared to their normally fluent peers.

Olander et al. (2010) used a synchronization-continuation task with clapping, comparing motor timing performance between CWS and CWNS ages 4–6 years. The children clapped to a beat for 12 beats (i.e., synchronization) and then continued the rhythm on their own for 32 beats (i.e., continuation). The children's performance in the continuation phase was analyzed. A relatively large subgroup (about 60%) of CWS demonstrated timing variability that was greater than the poorest performing child who did not stutter. However, the remaining CWS performed normally as compared with their peers.

Current Study

As a replication and an extension of the bimanual clapping study by Olander et al. (2010), we report analyses of the clapping abilities of 115 CWS and CWNS aged 3 to 9 years. An important goal of research on stuttering in childhood is to find predictors of ultimate persistence or recovery that can be used with preschool children to aid in decisions about early intervention. Therefore, we also searched for a relationship between stuttering persistence or recovery and earlier timing performance. Olander et al. (2010) assessed a sample of 17 CWS and 13 controls using data obtained in their initial year of testing in our longitudinal, 5-year project. In this investigation, we utilize data from multiple years of testing from a larger sample of 70 CWS and 45 CWNS who were followed for up to 5 years. The experimental task and dependent variables are identical to those reported by Olander et al. (2010). We used a bimanual clapping task with a synchronization-continuation paradigm. Clapping was chosen due its appropriateness for children as young as 4 years. With maturation, children become less variable in their clapping patterns with better motor coordination, reflecting the development of interlimb coordination (Fitzpatrick, Schmidt, & Carello, 1996; Getchell & Whitall, 2003). Thus, we expected both CWS and CWNS to improve on the clapping task in subsequent years of testing.

Based on the previous results from Olander et al. (2010), we expect that a motor timing deficit will characterize a subset of CWS. Last, because earlier investigations have reported differences in subgroups of children on speech and other motor abilities related to the following factors: sex (Smith & Zelaznik, 2004; Walsh, Mettel, & Smith, 2015), presence of language impairment (Brumbach & Goffman, 2014), and/or presence of phonological impairment (Ramus, Pidgeon, & Frith, 2003), we analyzed clapping timing performance of subgroups of children sorted on these variables.

Method

This report includes a replication and an extension of an earlier investigation from our laboratory, a cross-sectional study of bimanual clapping in preschool CWS and CWNS (Olander et al., 2010). Participants in both studies were recruited as part of the ongoing Purdue Stuttering Project. All recruitment, general testing, and data collection and analysis procedures were identical across the two studies. The present report contains a relatively large *n* cross-sectional comparison of clapping performance of 4- and 5-year-old CWS and CWNS in their first year of participation in the project. We also include observations derived from longitudinal data. Children made subsequent visits to the laboratory at yearly intervals. In the overall project, children participated in first sessions at different ages, from 3;0 (years;months) to 6;11. Children not only entered the project at varying ages, they also left the project at varying ages. As a consequence, the number of years and the age span available for longitudinal data analysis varied among the children. Because there are such rapid motor developmental changes over preschool and school-age periods (e.g., Getchell & Whitall, 2003; Smith & Zelaznik, 2004), all cross-sectional comparisons we implement use age-matched groups. Data collected in subsequent years of testing are used for two analyses in the present report: (a) cross-sectional comparisons of clapping performance using data from subgroups of children at each age from 3;5 to 9;5 years, and (b) tracking individual growth curves in clapping performance to ascertain if CWS and CWNS differ on the rate of acquisition of clapping skills. The longitudinal nature of this project allowed us to assess eventual persistence or recovery from stuttering in the later, school-age years (for those children followed long enough). Therefore, in the present report we also present clapping data derived from subgroups of CWS—those who eventually persisted and those who recovered.

Participants

Data from 115 children (70 CWS and 45 CWNS; aged 3;5 to 9;5) were used in the current study.¹ A diagnosis of stuttering was determined according to the criteria of Ambrose and Yairi (1999). These were (a) the child was regarded by the parent as having a stuttering problem; (b) the child was regarded by the project speech-language pathologist (SLP) as stuttering; (c) stuttering severity was rated as 2 or higher on an 8-point scale (0 = *normal* to

¹The data presented are a subset collected on our project examining multiple factors in stuttering. Many other tasks were included in project testing sessions, including electroencephalographic (EEG)/event-related potential (ERP) protocols to assess language processing, oral kinematic recording during speech production tasks, and electromyography (EMG) recorded during conversational speech. Identical testing procedures were performed at two locations, Purdue University and the University of Iowa. Note that data from the 30 children aged 4;0 to 6;10 used in the earlier investigation (Olander et al. 2010) were also used in the appropriate groups in the current study.

7 = severe) by the project clinician and the parent; (d) the child produced three or more stuttering-like disfluencies (SLDs)/100 syllables of spontaneous speech. The SLDs were coded from two spontaneous speech samples according to the methods of Ambrose and Yairi (1999). All coding of normal and stuttering-like disfluencies was completed by the project SLP and an SLP student trained by her. Student coders were trained on identifying and classifying disfluencies, and after training, their reliability was checked against that of the project SLP. All students achieved reliability >90% with the project SLP before undertaking coding of the conversational sessions. Recovery from stuttering was defined as 2 consecutive years in which the child did not meet the criteria listed above for a diagnosis of stuttering. Of the CWS, 30 children recovered and 29 children persisted in stuttering. Persistence or recovery status was not available for 11 children.

Large-Scale Cross-Sectional Analysis Ages 4;0 to 5;11

This analysis was implemented as a replication study of Olander et al. (2010) in which the mean age of the subjects was 5;0. Children were included in this analysis if they were between the ages 4;0 and 5;11 when first tested on the clapping paradigm. This resulted in groups of 47 CWS and 37 CWNS (see Table 1). The mean age was 4;6 for the CWS and 4;9 for the CWNS.

Multiyear Cross-Sectional Analysis of All Children

For the analysis over later years of clapping performance, children were grouped by age: 3;5–4;5 ($n = 27$), 4;6–5;5 ($n = 67$), 5;6–6;5 ($n = 78$), 6;6–7;5 ($n = 67$), 7;6–8;5 ($n = 52$), and 8;6–9;5 ($n = 23$). Overall, a total of 115 subjects were included in this analysis, comprising 70 CWS and 45 CWNS. Table 2 includes the breakdowns for each age group for CWSp, CWSr, and CWNS, in addition to the sex ratios for CWS and CWNS. As noted in the introduction to the Method section, n s at each year are not equal because children entered and left the project at variable ages (3–6 years) and left the project after a variable number of years (1–5). As shown in Table 2, the CWS were further subdivided on the basis of their final stuttering status, if known, when they left the project. Thus, these subgroups for the earlier years are retrospectively formed.

Table 1. Description of subjects in cross-sectional analysis between 4- and 5-year-old children who stutter (CWS) and children who do not stutter (CWNS).

Initial year	CWS	CWNS	Total
n	47	37	84
Average age in months (mean, median)	(55, 55)	(57, 57)	(56, 56)
Male:female	33:14	24:13	57:27

Note. Children were included in this analysis if they were between the ages 4;0 (years;months) and 5;11 when first tested on the clapping paradigm. Total n is less than 115 because some children were not in this age range when enrolled in the study.

Table 2. Description of subjects in analysis grouped by age for children who stutter who eventually persisted (CWSp), children who stutter who eventually recovered (CWSr), and children who never stuttered (CWNS).

Across years	3;5–4;5	4;6–5;5	5;6–6;5	6;6–7;5	7;6–8;5	8;6–9;5
n	27 ^a	67 ^b	78 ^d	67 ^d	52	23 ^d
CWSp	6	15	17	18	10	5
CWSr	9	21	23	21	17	8
CWNS	9	30	38	28	25	10
Male:female CWS	11:7	27:12	28:13	28:11	2:1	10:3
Male:female CWNS	7:2	9:5 ^c	22:13 ^e	19:9	14:11	1:1

Note. Children entered and left the project at varying ages depending on stuttering onset and referral, resulting in a varied number of years and age span available for data analysis at each year of age.

^aPersistence/recovery status was not included for three subjects.

^bPersistence/recovery status was not included for two subjects.

^cSex information was not included for two subjects. ^dPersistence/recovery status was not included for one subject. ^eSex information was not included for three subjects.

Screening/Testing Procedures Completed at Initial Recruitment

All of the children spoke American English as their first language. A pure tone hearing screening (20 dB HL at 400, 1000, 2000, and 4000 Hz) indicated normal hearing for all subjects. No motor delays, neurological problems, or serious illnesses were reported. A Handedness Inventory (subset of five tests adapted from Oldfield, 1971) indicated 86% of CWS and 80% of the CWNS were right-handed, whereas 4% of CWS and 13% of CWNS were left-handed. Handedness for the remaining percentages of each group was unknown. The general populations of CWS and CWNS participating in our project were matched on socioeconomic status, as determined by the mother's highest year of education (4 = high school graduate, 5 = partial college, 6 = college grad, and 7 = postgrad work; Hollingshead, 1975). The mean socioeconomic status score for each group was 6.

A set of standardized tests was administered to determine performance in a number of domains. Oral-motor and cognitive abilities were assessed through the administration of an oral-motor mechanism exam, the Columbia Mental Maturity Scale, and the Auditory Number and Word Memory subtests of the Test of Auditory-Perceptual Skills (Burgemeister, Blum, & Lorge, 1972; Gardner, 1996; Robbins & Klee, 1987). Both CWS and CWNS showed age-appropriate skills on these tests. To assess language and phonological status we administered the following tests: the Bankson Bernthal Test of Phonology (Bankson & Bernthal, 1990), the Test of Auditory Comprehension of Language—Third Edition (Carrow-Woolfolk, 1999), and the Structured Photographic Expressive Language Test—Third Edition (Dawson, Stout, & Eyer, 2003). When

compared with same-age peers, the CWNS who scored ≤ 1 *SD* below the mean for these test were excluded. CWS who did not pass these screening limits for phonology and language were not excluded. Thirty CWS scored ≤ 1 *SD* below the mean for phonology, and 13 scored ≤ 1 *SD* below the mean for language abilities. We included these children due to the high rates of co-occurrence of speech and language disorders in CWS (Arndt & Healey, 2001). Thus, our pool of CWS reflects the true heterogeneity of developmental stuttering.

Apparatus

A Northern Digital Optotrak 3020 (Northern Digital, Waterloo, Ontario, Canada) system was used to record index finger movements during the clapping task. The system consisted of three fixed cameras that tracked the motion of two infrared light emitting diodes (IRED) attached to the children's hands. The IREDs were connected to a small splint that was taped onto the distal end of each middle finger. The splint allowed the diodes to remain in view of the camera for the entire clapping motion. To avoid interference with clapping, wires were taped to the children's hands. IREDs were sampled at 250 Hz.

Procedure

The procedure we used is identical to that described in Olander et al. (2010). When the children were situated in front of the cameras with the IREDs attached, they were instructed to clap to a beat. The metronome beat consisted of a computer-generated piano tone (20 ms duration, 800 Hz) with an interbeat interval of 600 ms. After the synchronization phase of 12 beats, the metronome stopped, and children attempted to continue clapping as if the metronome were still on. The continuation phase continued for 32 claps until the children were instructed to stop. To ensure that children understood the task, up to three practice trials were completed before data collection began, with the experimenter clapping along with the child on the first practice trial. Participants were encouraged to complete six trials.

Kinematic Data Analysis

Because the clapping movement involved moving each hand toward and away from the midline in the transverse plane, the medial-lateral dimension from each IRED was analyzed (see the Appendix for details of kinematic data processing). A scoring algorithm implemented in a Matlab program and online examiner judgments were used to analyze the displacement data. Trials were excluded if the child stopped clapping for 2 s or more during the trial. It should be noted that for each trial, only the continuation phase of clapping was analyzed. Although the paced portions of the record potentially would yield interesting data concerning audio/motor integration skills, there were insufficient claps in the pacing interval to allow meaningful analyses. From the continuation phase, the first two claps and the last clap were excluded from the analysis. A minimum of two

usable trials containing at least 28 claps was required for a subject's data to be entered into the analysis.

From the displacement traces we computed average clap cycle duration, timing accuracy (root mean square error [RMSE]), and the coefficient of variation (CV) of interclap intervals within the time series for each trial for each child's left and right hand. To account for any possible influences of drift on average clapping rate, detrended variance was calculated, and its square root was used to calculate the CV in percent ($CV = [\text{Detrended standard deviation} / \text{mean interclap interval}] \times 100$). In timing studies, one is normally interested in timing variability of a clock-like process in which the clock setting (period) does not change. On some occasions a subject drifts off the prescribed interval goal. This drift adds a source of variability not of interest to the question at hand. By fitting the interval time series to a linear regression and then removing the linear trend, the remaining variability of the residuals are no longer influenced by a drifting clock-like process (see Robertson et al., 1999).

Results

Cross-Sectional Analysis of 4- and 5-Year-Old CWS Versus CWNS

Results are reported for 47 CWS and 37 CWNS who produced at least two useable clapping trials. The range of useable trials per child was two to 11 trials, with a median of six useable trials per child for both groups. Olander et al. (2010) addressed the statistical concern that clapping variability may be affected by the number of useable trials by computing the correlation between the number of useable trials and the CV of the interclap interval. The correlation was almost zero (-0.075), thus mitigating this concern.

Table 3 contains means and standard deviations for clapping cycle duration, CV, and RMSEs for stuttering and nonstuttering groups subdivided by sex. Repeated measures analyses of variance (ANOVAs) were computed on three dependent variables—cycle duration, CV, and RMSE—for the 4- and 5-year-olds' cross-sectional data set. In all

Table 3. Mean and standard deviation for clap duration (ms), detrended coefficient of variation (CV; %), and root mean square error (RMSE; ms) by sex and group for the initial cross-sectional groups.

Dependent variable	CWS male (n = 35)	CWS female (n = 14)	CWNS male (n = 24)	CWNS female (n = 13)
Duration (ms)	466.30 (75)	520.80 (73)	477.90 (61)	487.60 (79)
Detrended CV (%)	19.21 (9)	18.28 (8)	17.19 (7)	17.12 (8)
RMSE (ms)	153.70 (57)	110.29 (46)	132.80 (57)	134.70 (58)

Note. Data for RMSE were not included for seven male CWS, four female CWS, and eight male CWNS. CWS = Children who stutter; CWNS = Children who never stuttered.

three ANOVAs, hand was the within factor, and stuttering group and sex were between-subjects factors.

Figure 1 shows the distribution of mean clapping interval measured separately for the right and left hands for each child. As seen in this plot, no group differences between CWNS and CWS were found for average clap duration, $F(1, 79) < 1$. Sex did not have an effect on duration, $F(1, 79) = 3.2, p = .08$, nor was there a significant stuttering group by sex interaction, $F(1, 79) = 1.5, p = .22$. There was no effect of hand on cycle duration, $F(1, 79) < 1$, and no interaction of hand with stuttering group or sex.

Figure 2 contains a scatter plot of the CV of the interclap intervals for the right and left hands for each subject. From the plot, it is apparent that the distributions of the children's mean CV are overlapping for the two groups of subjects: stuttering and nonstuttering. There was no significant difference in CV for CWS and CWNS, $F(1, 77) < 1$, nor any effect of sex, $F(1, 77) < 1$, and no significant interaction between the factors, sex and group, $F(1, 77) < 1$. There was no effect of hand on CV, $F(1, 77) < 1$. Unlike the results observed in our earlier study in which 60% of CWS fell outside the CV range for clapping observed in the CWNS (Olander et al., 2010), CWS in the current study did not split in two performance groups. There was not a subgroup of CWS who performed remarkably poor or well.

Figure 3 shows accuracy of clapping performance, calculated as RMSE, for the right and left hands. There was no significant difference between RMSE for CWS and CWNS, $F(1, 58) < 1$. Sex did not have an effect on interclap interval variability $F(1, 58) = 1.95, p = .17$. As for cycle duration and CV above, hand had no effect on RMSE $F(1, 58) < 1$, and there was no significant interaction of hand with the either of the two between groups variables.

Last, Olander et al. (2010) speculated that the large percentage of CWS who performed so poorly on timing that their CVs fell outside the range of the CWNS would be more likely to have a persistent stuttering problem. We did not have persistence/recovery data at the time of that report. That hypothesis can be addressed now by retrospective analysis of the data from the larger pools of 4- and 5-year-old children in our current report, whom we have followed for up to 5 years. Therefore, the circles representing CWS in Figures 2 (CV) and 3 (RMSE) have been subclassified as persistent (red) or recovered (green). Examination of the distribution of the data for CWS_p and CWS_r helps to answer the question of whether the CWS who would eventually persist in stuttering (CWS_p) in the school-age years were among the most variable in clapping timing performance when they were preschoolers. It is clear that the clapping variability of the CWS_p is distributed along the entire continuum from the best to the worst timers when estimated both by CV and RMSE measures. In fact, there is a significant cluster of CWS_p with extremely consistent (e.g., CVs in the 10% or lower range) clapping performance.

Phonological and Language Status

Groups of CWS with language and/or phonological delay/impairment (those who scored below 1 *SD* below the mean on a standardized language or phonological assessment) were analyzed separately to examine whether a lag in one of these areas had an effect on clapping performance. Table 4 contains mean and standard deviation values for each group for clapping cycle duration and CV. RMSE measures are not reported because we were missing RMSE data for 19 subjects. Comparing the means of each group relative to their associated standard deviations suggests that, similar to stuttering status, neither language nor phonological

Figure 1. Cycle duration (ms) for children who stutter (CWS) and children who do not stutter (CWNS) in the initial year of the study (target 600 ms). No group differences were found between CWS and CWNS for cycle duration.

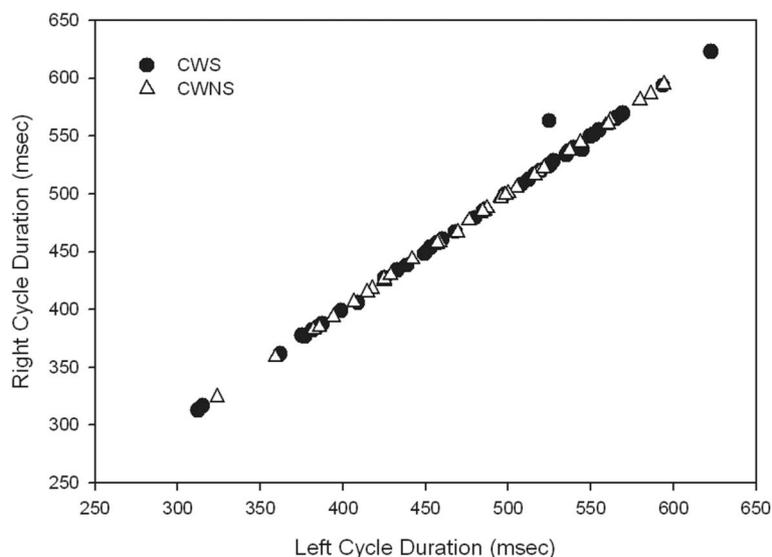
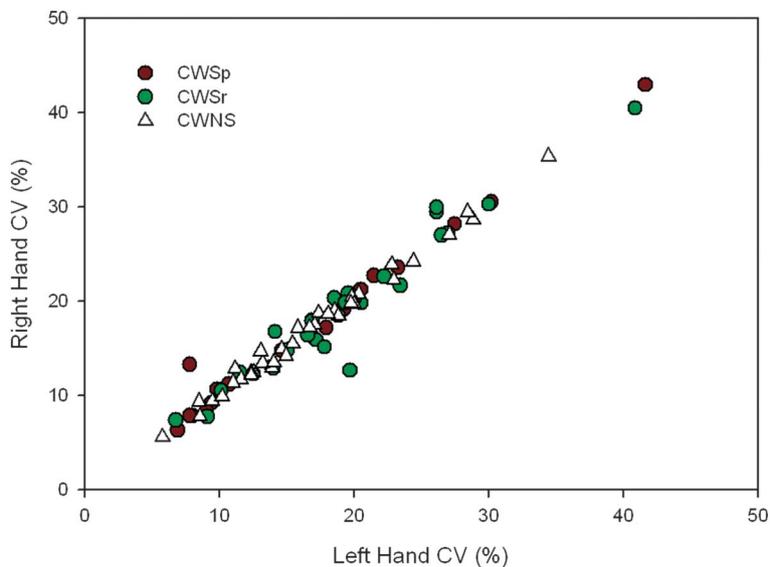


Figure 2. Mean detrended coefficient of variation (CV; %) of interclap intervals for 4- to 5-year-old preschool children who stutter who eventually recovered (CWSr; green), children who stutter who eventually persisted (CWSp; red), and children who never stuttered (CWNS; open triangle). The poorest performing children include children from all three groups, and no group differences were found.



delay or impairment has a significant impact on mean clapping rates or variability in maintaining the beat.

Analysis of Clapping Longitudinally

Results are reported for 115 subjects grouped into CWSp, CWSr, and CWNS. Data from children followed

for up to 5 years were organized into six age groups. The children entered and left the study at varying ages depending on stuttering onset and study referral, accounting for the differences in sample size (see Table 2) across the age groups. Figure 4 shows the means for average clap duration for each group at each age (with a target interclap interval of 600 ms). The groups follow a similar trajectory with clap

Figure 3. Mean root-mean-square error (RMSE) for 4- to 5-year-old preschool children who stutter who eventually recovered (CWSr; green), children who stutter who eventually persisted (CWSp; red), and children who never stuttered (CWNS; open triangle). The poorest performing children include children from all three groups, and no group differences were found.

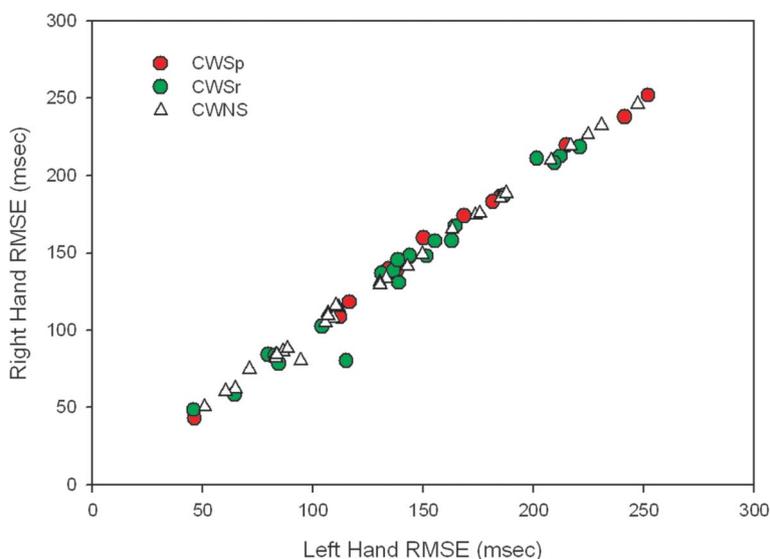


Table 4. Mean and standard deviation clap duration (ms) and detrended coefficient of variation (CV; %) by language impairment (LI) and stuttering group and phonological impairment (PI) group for the initial year of cross-sectional groups.

Language	CWS + LI (n = 8)	CWS – LI (n = 35)	CWNS (n = 37)
Duration (ms)	507.9 (74)	469.1 (76)	482.0 (66)
Detrended CV (%)	21.1 (7)	18.6 (9)	16.9 (7)
Phonology	CWS + PI (n = 24)	CWS – PI (n = 22)	CWNS (n = 37)
Duration (ms)	490.9 (67)	470.8 (86)	482.0 (66)
Detrended CV (%)	20.5 (9)	17.3 (8)	16.9 (7)

Note. CWS = Children who stutter; CWNS = Children who never stuttered.

interval duration increasing closer to the target rate across the developmental continuum. There is no hint of any differences in clapping rate among the three groups of children, CWSp, CWSr, and CWNS at any age.

Figure 5 contains a plot showing the means for CV for each group at each age. For the oldest group, data from one CWSp were omitted, as his CV was an outlier (more than double that of any other child at this age). As with cycle duration, these plots indicate no differences among the three groups at each age sampled. These cross-sectional comparisons by year across development for CWSp, CWSr, and CWNS suggest no evidence that atypical basic manual timing control is an early factor in stuttering, nor that it emerges as a factor in the school-age years. Another way

to approach this issue is to ask whether there are CWS who show atypical individual growth curves in their development of basic manual timing control processes. To address that question, we plotted individual growth curves for each child for whom 2 or more years of clapping data were available. The individual growth curves in Figures 6A, 6B, and 6C further support the similarities in developmental profiles among CWSp, CWSr, and CWNS. Thus, the group of CWSp was not different in their mean rate of clapping nor were they more variable in maintaining the rate at any age. In addition, they did not differ in the maturational course of improving performance on this basic motor skill.

Discussion

Three notable findings emerge from our results: (a) our large-scale cross-sectional study of 4- and 5-year-old children did not reveal differences in clapping timing behavior between CWS and CWNS, nor was there a subgroup of CWS who were particularly poor at timing in the clapping task; (b) simple motor timing mechanisms as revealed in the bi-manual clapping task for preschoolers are not predictive of persistence or recovery from stuttering; and (c) the development of clapping skill improved over the school-age years in a similar manner in both children diagnosed as stuttering and those who have never stuttered.

With regard to the first finding, clearly our results do not replicate the findings reported by Olander et al. (2010). In the earlier study we found significant overall group differences between CWS and CWNS in the CV measure, an estimate of the consistency in maintaining the clapping

Figure 4. Mean clap interval (ms; error bars are SEM) as a function of age for children who stutter who eventually persisted (CWSp; red), children who stutter who eventually recovered (CWSr; green), and children who never stuttered (CWNS; open triangle) across six ages. The target rate for the interbeat interval was 600 ms. The groups follow a similar trajectory with clap interval duration (Dur) increasing closer to the target rate with maturation. yr;mo = year;month

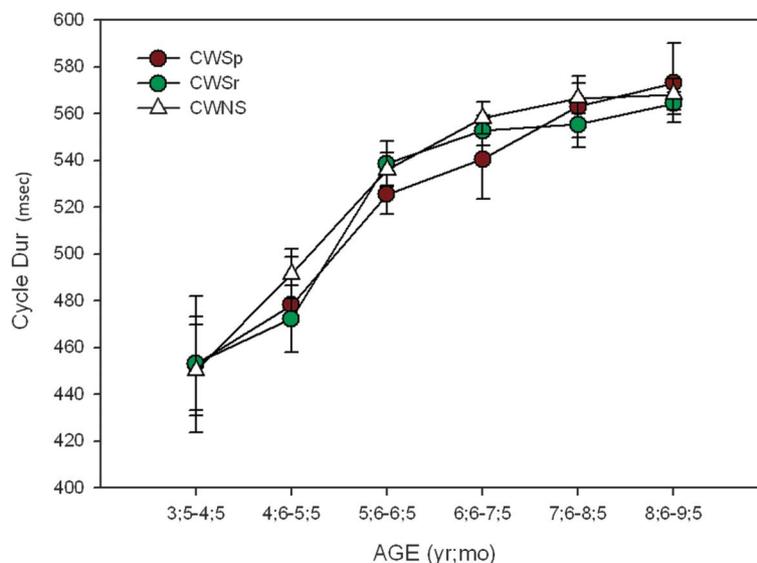
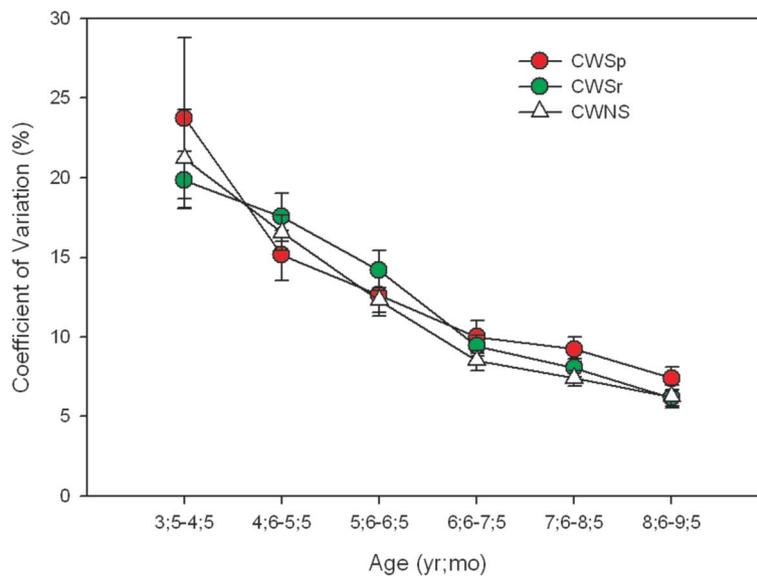


Figure 5. Mean detrended coefficient of variation (CV; %; error bars are *SEM*) for interclap intervals as a function of age for children who stutter who eventually persisted (CWSp; red), children who stutter who eventually recovered (CWSr; green), and children who never stuttered (CWNS; open triangle) across six ages. yr;mo = year;month



interval. Olander et al. (2010) also found a subgroup of CWS, 60% of the 17 subjects, who performed outside the range of the typically developing children. It is clearly critical to question why results of two studies from the same laboratory would yield such disparate results. Obvious factors to consider include methods of subject recruitment, data collection, and data analysis. Subject recruitment methods were identical to those used to recruit the earlier samples of CWS and CWNS. The populations from which the subjects are drawn have not changed. Methods for recruiting children, screening tests to qualify children as typically developing, and diagnostic criteria for stuttering were carefully and precisely prescribed at the beginning of the project, and they have been rigorously implemented and unchanged. Data collection procedures for the clapping protocol are identical to those used in the earlier phase of the project. Concerning kinematic data scoring for the clapping analysis, our methods are identical to those used with the smaller groups of subjects whose data were included in the earlier report. It is of importance that the methods for computing each subject's CV are unchanged. There was a change in our choice of an error measurement. In the present report we use RMSE, but similar to the phase analysis we used earlier, no significant or interesting results arose from measure.

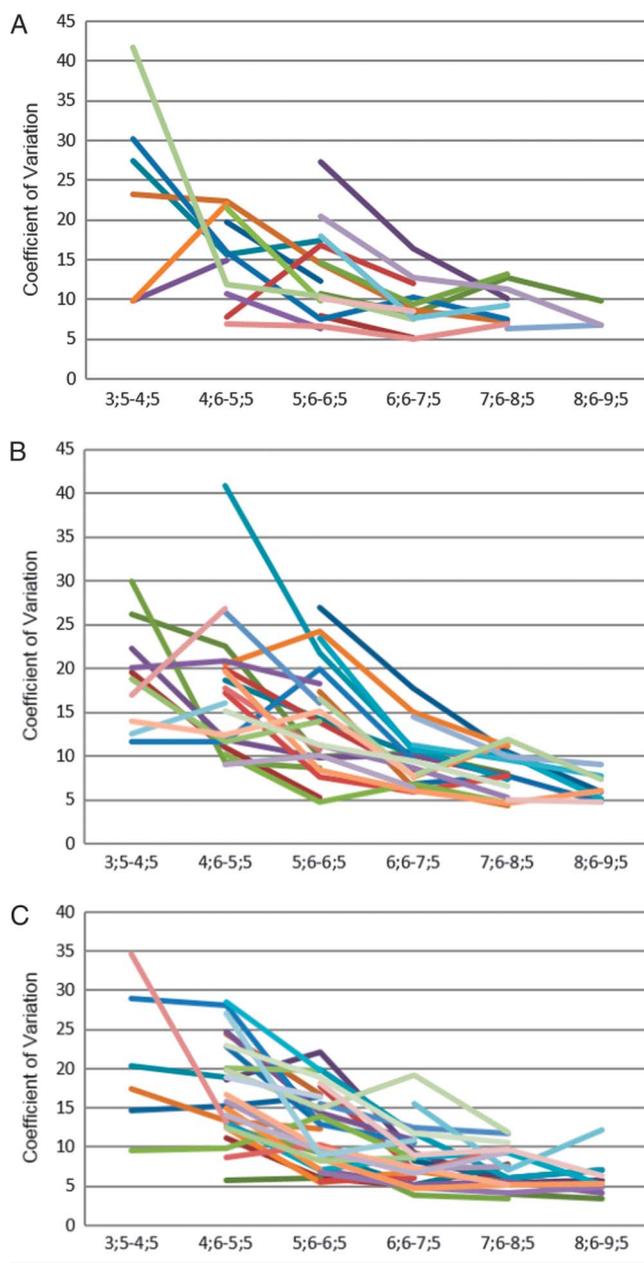
Ruling out the methodological factors mentioned above, we argue that the most likely explanation for the differences in results between Olander et al. (2010) and the present report is the difference in sample sizes. It seems highly likely that using the relatively small sample of 13 CWNS, whose data formed the basis for the earlier report, resulted in a significant underestimate of the true range of the CV of clapping in the population of nonstuttering preschool

children. Table 5 reports the observed ranges of clap interval and CV of clap interval from the present study and from the earlier study for the two groups of children. In the earlier study (Olander et al., 2010), data from the 13 CWNS produced a slightly reduced clapping interval range, compared to the present report; however, the range of the CV of clapping, which was only 5%–12%, is remarkably reduced compared to the range of CV, 5%–35%, in the present report. Thus it seems likely that random sampling error produced an estimate of the range of typical clapping consistency for the nonstuttering preschool population that was much lower than the true population value. Therefore the preschool CWS whom we reported to perform outside the normal range of clapping abilities for their age were actually well within the typical range for this age.

Regardless of whether there are differences between the means of the two groups, stuttering versus nonstuttering, on a specific variable, one can still ask whether that variable is predictive of persistence or recovery. In other words, are preschool CWS who are performing on the low range of timing control in the clapping task more likely to persist in stuttering? The answer is no. When 4- and 5-year-old children's CV measures from their initial testing on the clapping paradigm were retrospectively classified according to eventual stuttering status—never stuttered, recovered, or persisted—there was no suggestion that the CWSp performed more poorly at age 4 to 5 years. CWSp were among the most variable timers and the least variable timers in the clapping task (see Figure 2).

Our longitudinal analyses also revealed no remarkable differences between the groups of children classified according to eventual stuttering status. All three groups of children showed dramatic improvements in clapping closer

Figure 6. Individual detrended coefficient of variation (CV) data for clapping performance in children who stutter who eventually persisted (CWSp; A), children who stutter who eventually recovered (CWSr; B), and children who never stuttered (CWNS; C). Each subject is represented by a different color line. The individual profiles further exemplify the similarities observed across the three groups.



to the actual target rate (interclap interval of 600 ms) and in consistency of maintaining the beat as indexed by the CV of clapping (see Figures 4 and 5). Furthermore, we found no evidence that CWSp showed different individual growth curves in their clapping abilities (see Figure 6).

An obvious question that arises from the earlier mixed results of studies of nonspeech motor abilities of AWS is whether a different, perhaps a more complex, task would

Table 5. Comparison of results from Olander et al. (2010) and the present study. CWNS = children who do not stutter; CWS = children who stutter.

Group (n)	Clapping interval range (ms)	Mean (ms)	Coefficient of variation range (%)	Mean (%)
Olander et al., 2010				
CWNS (13)	375–600	464	5–12	8.9
CWS (17)	300–500	427	5–29	14.2
Present report				
CWNS (37)	325–600	482	5–35	16.9
CWS (47)	300–620	493	5–42	18.9

have revealed differences between CWS and CWNS and/or contribute to predicting persistence/recovery. This certainly seems to be a possibility, but in assessing preschoolers, the experimenter is limited by the capabilities of the young child. Bimanual clapping seems an ideal task, because it is familiar—children clap in everyday life—and it involves intereffector coordination, which the earlier literature suggests might be critical for finding stutter/nonstutter differences (Zelaznik et al., 1997).

One interesting finding of the current study is that the youngest children, for the most part, clapped at a faster rate during the continuation phase, approximately 400–500 ms, than the target interbeat interval, 600 ms, that they heard in the synchronization phase. We chose this target rate because we did not want the task to be too difficult for the preschoolers, and we believed a relatively slow rate would be easier. With hindsight, we might posit that a faster target rate would be easier for 4- and 5-year-olds. A study of spontaneous finger tapping rates adopted by children and adults across the life span (McAuley, Jones, Holub, Johnston, & Miller, 2006) demonstrated that children aged 4–5 years produce preferred rates with interbeat intervals of about 200–500 ms. Therefore it is possible that the youngest children in our study produced rates during the continuation phase that were closer to their naturally preferred tempos. The improvement toward the 600-ms target interval observed in the older groups of children (e.g., Figure 5) also is consistent with the findings of this study in that older children aged 8–9 years had slower preferred tempos, about 400–600 ms.

An earlier study of older and younger groups of school-age children (Falk et al., 2014) reported differences in stuttering and nonstuttering groups in a task requiring synchronization of finger tapping to an external beat. Therefore, another obvious experimental direction would be to analyze synchronization abilities of preschool children, rather than the continuation phase following a brief synchronization period as we elected to do. Analyzing the synchronization behavior would tap into auditory/motor integration, which is implicated in stuttering (e.g., Cai et al., 2012). Given that the synchronization phase we used was so brief, only 12 beats, the available data would not yield meaningful results. Future studies of longer synchronization trials perhaps with rhythms that vary from simple to more complex would be useful to answer this question.

Conclusion

The present report comprises, to our knowledge, the most in-depth and relatively large cross-sectional and longitudinal study of basic motor timing abilities in CWS. We find no evidence that the neurodevelopmental processes involved in the early course of stuttering include atypical growth of basic motor timing networks used in nonspeech motor behaviors. We have suggested above that other motor tasks could reveal atypical functions; however, our two studies, taken together (Olander et al., 2010, and the present report) provide a clear warning about drawing conclusions about stutter/nonstutter differences on the basis of data derived from relatively small samples. Most previous studies of nonspeech motor abilities of AWS and CWS have used group sizes typically in the range of eight to 15 subjects.

Another important point to consider when interpreting current findings is that the children in our project participated in multiple data collection protocols. Although we found no differences between them in the clapping task, there are significant differences between the stuttering/nonstuttering preschool groups on event-related potentials (ERPs) related to language processing (e.g., Weber-Fox, Wray, & Arnold, 2013) and in indices of speech motor control and coordination (e.g., Walsh et al., 2015). Therefore, early stuttering may be associated more specifically with atypical growth of neural networks supporting speech and language functions of the CNS.

Acknowledgments

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Appendix

Detailed Methods for Kinematic Analysis of Clapping Records (Adapted from Olander et al., 2010)

Displacement records for right and left hands were low-passed filtered (cut-off 8 Hz) in the forward and reverse directions. Motions of each hand were measured separately. The start point for each clap was defined as the point at which the velocity of the hand slowed to 3% of the peak velocity while moving towards the midline. This 3% velocity point towards the midline corresponds almost exactly to the point in time when the hands first made contact. The starting point of each clapping cycle also served as the ending point of the previous cycle. We used a Matlab algorithm to automatically detect the starting point for each clap using the 3% velocity criterion. The displacement of both the hands and the automatically defined claps were displayed graphically as the experimenter analyzed each clapping trial. If the automatic algorithm clearly picked an erroneous starting point, the user changed the point to the correct location using a cursor. Similarly if a clap was missed by the algorithm, it was added in the correct location by the user. We have used similar methods in previous studies of rhythmic movements (Robertson, 1999; Zelaznik et al., 1997).