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Research Note

Use of Automated Kinematic Diadochokinesis Analysis to Identify Potential Indicators of Speech Motor Involvement in Children With Cerebral Palsy

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ABSTRACT

Purpose: This study examined multiple variables obtained from an automated measure of lip movement during a diadochokinesis (DDK) task to identify those with potential to detect mild speech motor involvement in school-age children diagnosed with cerebral palsy (CP).
Method: Eight children with CP and high speech intelligibility and a matched group of eight children with typical development (TD) completed a DDK task while their lip and jaw movements were recorded. A custom MATLAB algorithm was used to automatically extract 23 kinematic measures of children's lip movements during production of the DDK sequences. Mann–Whitney *U* tests were used to compare groups on the kinematic measures, and receiver operating characteristic (ROC) analysis was used to evaluate the diagnostic accuracy of measures that significantly differed between groups.
Results: Five of the 23 kinematic variables differed significantly between the CP and TD groups. These were two measures of overall DDK performance (i.e., duration of the DDK sequence and number of cycles) and three spatial and temporal measures of lip movement. Duration of the DDK sequence and the mean displacement of the lips across cycles had the highest diagnostic accuracy, differentiating CP and TD groups with 88% sensitivity and 88% specificity.
Conclusions: Automatically derived kinematic measures of DDK sequences differentiated children with CP and high intelligibility from typically developing children. Future research is needed to determine the clinical utility of these measures for detecting speech motor impairment.

Cerebral palsy (CP) is the most common cause of motor disability in children, affecting approximately two to three per 1,000 children around the globe (Maenner et al., 2016; Sellier et al., 2016). Over 50% of children with CP have dysarthria (Cockerill et al., 2014; Nordberg et al., 2013), which may be characterized by reduced intelligibility (Allison & Hustad, 2018b; Hustad, Sakash, Broman, & Rathouz, 2019; Hustad, Sakash, Natzke, et al., 2019; Lee et al., 2014), slower speaking rates (Allison & Hustad, 2018a, 2018b; Hustad et al., 2010), reduced ability to manipulate pitch ranges (Kuschmann & Lowit, 2019), irregular rate and rhythm (Ansel & Kent, 1992), and disordered perceptual characteristics including hypernasality and atypical voice quality (e.g., Schölderle et al., 2020; Workinger & Kent, 1991).

Dysarthria is diagnosed through the identification of perceptual speech features (Darley et al., 1969; Schölderle et al., 2020) that are caused by neuromotor impairments to the speech mechanism (Darley et al., 1969). Although not all children with CP meet clinical auditory-perceptual criteria for a dysarthria diagnosis, they may still show subtle deficits in speech motor performance when compared with children with typical development (TD), including reduced interarticulator coordination (Nip, 2017), increased ranges of movement and decreased speech movement stability (Nip et al., 2017), and impaired vocal fold articulation (Nip

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& Garellek, 2021) in a range of speech and nonspeech tasks. Furthermore, at the auditory-perceptual level, children with CP but no clinical diagnosis of dysarthria still lag behind typically developing peers in intelligibility development (Allison & Hustad, 2014; Hustad, Sakash, Natzke, et al., 2019; Hustad et al., 2012). For instance, children with CP and no diagnosis of dysarthria can show intelligibility deficits when pushed to produce utterances longer than their typical utterance length (Allison & Hustad, 2014).

Collectively, these findings demonstrate that even children with CP who do not have dysarthria can exhibit difficulties with speech motor control when asked to perform tasks that challenge their motor capacity. Although children with CP who have high intelligibility may not need speech intervention, understanding how their speech motor control differs from children without CP may elucidate the underlying impact of CP on speech biomechanics. This knowledge will enhance understanding of the range of speech motor abilities in children with CP and may help identify core speech movement deficits that could be potential targets of intervention for children with CP who do have dysarthria. In addition, identifying objective measures and speech tasks that are sensitive to speech motor deficits in children with a known motor disorder but high intelligibility may eventually lead to improved methods for detection and differential diagnosis of dysarthria in pediatric populations.

Diagnostic Utility of Maximum Performance Tasks

Maximum performance tasks, for example, oral diadochokinesis (DDK) and maximum phonation duration, have been shown to distinguish children with speech motor impairments from typically developing peers (Thoonen et al., 1996, 1999; Wit et al., 1993). Although these tasks are not true speech tasks (e.g., Ballard et al., 2009; Schölderle et al., 2018), the underlying premise of maximum performance tasks is that they are designed to examine the limits of an individual's speech motor capacity (Nip & Green, 2013; Rong et al., 2018). For example, a maximum performance task such as a maximum phonation duration task can be used to provide information about the motor capacity of respiratory control and efficiency of laryngeal valving.

Examining maximum performance tasks produced by children with dysarthria, children with childhood apraxia of speech (CAS), and children with TD, Thoonen et al. (1996, 1999) determined that using tasks such as DDK and maximum phonation duration could distinguish these children with high degree of sensitivity and specificity. This finding suggests that each group (CP, CAS, and TD) have differing profiles in their speech motor capacity. Quantitative measures of DDK performance do not provide the comprehensive clinical information needed for diagnosis and treatment decision making (e.g., about the child's auditory-perceptual speech features or intelligibility); however, they may be an efficient clinical tool to evaluate the speech motor capacity of individuals with suspected speech motor impairments. In comparison with maximum phonation duration, which primarily engages the respiratory and laryngeal subsystems, DDK requires respiratory, laryngeal, velopharyngeal, and articulatory subsystem involvement. In the clinical administration of DDK, children are typically asked to repeat a syllable, such as "pa" or "ba," as quickly as possible and for as long as possible (Rvachew et al., 2006; Schölderle et al., 2018; Thoonen et al., 1996). This task theoretically requires the child to maximize their respiratory support, maintain a consistent subglottal pressure (combination of respiratory and laryngeal subsystems), maintain consistent vocal fold adduction, maintain consistent velopharyngeal closure, and quickly alternate articulatory movements (e.g., closing and opening the lips) while building adequate intraoral pressure for the consonant. DDK can be further increased in difficulty by including consonant sequences (e.g., "pataka") to require alternation between multiple places of articulation. Because multiple speech subsystems are often affected in children with dysarthria (Allison & Hustad, 2018a; Lee et al., 2014; Nip, 2017; Nip & Garellek, 2021), DDK tasks that challenge their speech motor abilities at multiple subsystem levels might be particularly sensitive and specific in detecting speech motor impairments.

Kinematic analyses of DDK, therefore, may be an efficient way to determine whether a child has speech motor difficulties. However, aside from a finding that spatial and temporal interarticulator coordination is reduced for children with CP as compared with their TD peers for DDK (Nip, 2017), no between-groups study has examined the kinematic characteristics of DDK. Improved understanding of how oral movements during DDK differentiate children with CP from their TD peers would provide insights about how the speech motor system is affected in this disorder and inform development of objective markers for identification of speech motor impairment in children. Although other more naturalistic speech tasks, such as production of longer sentences, could also challenge children's speech motor systems and potentially reveal subtle speech motor involvement, the DDK task has several unique advantages pertaining to its simple phonological structure, which allows the task to be easily administered in pediatric populations. DDK also enables automated, objective measurements of spatial and temporal dimensions of speech movement that cannot be easily obtained from sentence production. This study extends prior work of Thoonen et al. by investigating the ability of novel kinematic measures of DDK performance to differentiate children with CP from neurotypical children.

Algorithmic Approaches to Analyzing Kinematic Data

Recently, Rong et al. (2018) developed an algorithm to automatically extract 22 kinematic variables from DDK tasks. These variables examined spatial (e.g., range of movement), temporal (e.g., duration), and spatiotemporal (e.g., speech movement variability and velocity) aspects of articulatory movements during a DDK task (Rong et al., 2018). Some of these kinematic variables provide metrics of the entire sequence, including duration, velocity, and the number of cycles produced. The algorithm also parses the individual syllable repetitions within the DDK trial and provides metrics of variability across cycles.

Rong et al. (2018) found the validity of the automated algorithm to be high as it was strongly correlated ($r > .99$) with manual measurements of the same variables. In addition, the algorithmically derived variables were able to distinguish people with amyotrophic lateral sclerosis from neurologically healthy peers (Rong, 2020; Rong et al., 2018). Applying this approach to children with CP may lead to new insights about how the disorder affects articulation and speech motor development in this population and help to construct etiology-specific articulatory profiles to inform and guide clinical management.

Research Questions

The aim of this preliminary study was to determine which aspects of articulatory movements differ between children with CP and children with TD using Rong et al.'s (2018) automated algorithm. Our research questions for this exploratory study were the following:

(1) What spatial, temporal, and spatiotemporal aspects of articulatory movements during DDK production distinguish children with CP with high intelligibility from their TD peers?

(2) What are the sensitivity and specificity of the spatial, temporal, and spatiotemporal aspects of oral movements during DDK production that distinguish children with CP with high intelligibility from their TD peers?

Method

Participants

Sixteen school-age children participated in this study: eight children with a medical diagnosis of spastic CP and eight children with TD. Data for this project were collected at two different sites (MGH Institute of Health Professions and San Diego State University [SDSU]) as part of separate ongoing projects (Allison et al., 2020; Nip et al., 2017). Participant details are shown in Table 1.

Children with CP were four males and four females between ages 7–15 years (M = 10.9 years, SD = 2.8 years). Because the purpose of this study was to examine objective speech measures that are sensitive to mild speech motor

Table 1. Demographic characteristics of children in the cerebral palsy (CP) and typical development (TD) groups.

Group	Child ID	Age (years)	Gender	Intelligibility (%)	GMFCS	Anatomic involvement	Type of CP	Auditory perceptual speech features	Dysarthria present?
CP	CP1	7.4	F	86.0	II	Diplegia	Spastic	Reduced pitch, mild strained-strangled voice, hypernasality, glottal fry	Yes
	CP2	8.6	M	95.4	I	Left hemiplegia	Spastic	Hoarse vocal quality, monopitch, imprecise articulation, vowel distortions	Yes
	CP3	9.0	M	96.0	II	Diplegia	Spastic	Mild /s/ articulation error	No
	CP4	10.2	F	98.0	III	Unknown	Spastic	Hyponasality, reduced pitch	Yes
	CP5	10.6	M	96.0	IV	Quadriplegia	Spastic	/r/ articulation error	No
	CP6	12.3	M	96.0	II	Diplegia	Spastic	Some glottal fry	No
	CP7	14.6	F	99.2	III	Diplegia	Spastic	Glottal fry, imprecise articulation	Yes
	CP8	15.0	F	93.0	II	Diplegia	Spastic	None	No
TD	TD1	7.3	F	88.0					
	TD2	8.6	M	97.9					
	TD3	9.0	M	92.0					
	TD4	10.8	F	98.0					
	TD5	10.9	M	96.0					
	TD6	12.3	F	99.6					
	TD7	13.2	M	99.0					
	TD8	15.6	F	98.0					

Note. GMFCS = Gross Motor Function Classification System level (Palisano et al., 1997); F = female; M = male.

involvement, children with CP were selected for inclusion who had high speech intelligibility. Children with CP whose intelligibility in sentences on the Test of Children's Speech+ (TOCS+; Hodge & Daniels, 2007) was over 85%, based on orthographic transcription by naïve listeners, were specifically selected for this study. Procedures for measuring intelligibility have been fully described in previous publications (Nip, 2017; Nip et al., 2017). Briefly, children repeated sets of sentences between two and seven words in length from the TOCS+, totaling 80 words per child. Three unique unfamiliar listeners listened to recordings of each child's sentence productions and orthographically transcribed what they thought the child said. The percent of words correctly identified by each listener was calculated. For each child, scores were averaged across the three listeners to yield their percent intelligibility score.

The children with CP all participated in a speech and language assessment, including speaking tasks of varying length and complexity. Three expert speech-language pathologists (SLPs) independently rated the presence of auditory-perceptual features of dysarthria for each child, as a basis for determining a dysarthria diagnosis. SLPs listened to a connected speech sample from each participant (i.e., a conversation sample from the SDSU participants; production of the TOCS+ sentences for the MGH participants) and judged whether any of the widely used Darley et al. (1969) dysarthria characteristics were present in each speech sample. The SLPs then compared their judgments, discussed any disagreements until a consensus was reached about the characteristics exhibited by each child, and determined whether the child had dysarthria based on their speech characteristics. Four of the children with CP met criteria for dysarthria, and four were determined to not have dysarthria. Auditory-perceptual speech characteristics and dysarthria diagnosis for each child are listed in Table 1. Seven out of the eight children with CP exhibited some atypical speech characteristics for their age, even if they did not meet criteria for dysarthria. Because the study aimed to identify objective measures sensitive to subtle speech motor involvement, children were selected for inclusion based on their intelligibility, rather than the presence or absence of dysarthria.

All children with CP passed a hearing screening at 0.5, 1, 2, and 4 kHz and achieved scores in the average range on standardized language testing (i.e., Clinical Evaluation of Language Fundamentals–Fourth Edition [CELF-4] Core Language Scale [Semel et al., 2003], SDSU participants) or passed a language screener (i.e., CELF-4 screening test [Semel et al., 2004], MGH participants). Gross motor skills and anatomic involvement varied, but all participants had been medically diagnosed with spastic CP. Children with TD were matched for age and gender to the children with CP (M_{age} = 10.9 years, SD = 2.7 years). All children in the TD group passed speech, language, and hearing screenings and had no reported history of developmental delay or other childhood neurological conditions.

Procedure

Speaking Task

All children performed a DDK task in which they were asked to produce the syllable "ba" as quickly and accurately as possible on one breath. "Ba" was used rather than other DDK tasks such as "ta" or "pataka," because bilabial consonants were needed for kinematic analysis of sequences. Although "pa" is the bilabial syllable more frequently used in clinical DDK assessment, DDK in this project was collected as part of a research protocol in which production of "ba" was compared across speaking tasks (Nip, 2017). Audio and kinematic data were simultaneously recorded during the task. Children were provided with instructions, a verbal model by the examiner and one to two practice trials of the task. Every child produced at least two DDK sequences following the practice items. For each child, the first sequence in which they produced at least 10 consecutive repetitions of "ba" was analyzed using the automated DDK analysis. Two children in the CP group (CP7 and CP8) were only able to produce a maximum of five syllables on one breath across all DDK trials; thus, analyses for these children are based on their longest DDK sequences (i.e., five syllables). Within the CP group, the number of cycles produced in the analyzed DDK sequence ranged from five to 38 across children. In the TD group, the number of cycles analyzed ranged from 19 to 77.

Kinematic Recording

Children's lip and jaw movements were recorded during the DDK task. At one data collection site (SDSU), a three-dimensional optical motion capture system (Motion Analysis, Inc.) was used to record the movement of the upper lip, lower lip, and jaw during DDK production at a sampling rate of 120 Hz. Data from six of the children with CP (CP1, CP3–CP6, and CP8) and six of the children with TD (TD1, TD3–TD5, and TD7–TD8) were collected at this site. At the second site (MGH IHP), electromagnetic articulography (EMA; Wave, Northern Digital) was used to record kinematic data at a sampling rate of 100 Hz. Data from two of the children with CP (CP2 and CP7) and two of the children with TD (TD2 and TD6) were collected at this site. Standard marker placements on the upper lip (UL), lower lip (LL), and jaw (J) were used across sites. The motion capture and EMA systems have comparable measurement precision (the accuracy and precision of both systems are 0.1–0.6 mm; Motion Analysis Ltd., Northern Digital; Berry, 2011); thus, the kinematic data can be combined for analysis.

Automated DDK Analysis

All movement time series from the lips and jaw were corrected for head movement. An automated DDK MATLAB program (Rong et al., 2018) was used to analyze kinematic data from DDK sequences. Because we focused on a DDK sequence involving bilabial consonants, the UL–LL distance signal was used for analysis. The procedures of the automated kinematic DDK analysis were described in the work of Rong et al. (2018). Briefly, the UL–LL distance time series for each child was semiautomatically parsed into cycles that corresponded to individual syllable repetitions. On the basis of the parsed and complete lip kinematic data for the DDK sequence, 22 variables that characterized the spatial, temporal, and spatiotemporal features of lip movement were automatically extracted for each child using an algorithmic approach. Because kinematic measures were algorithmically determined, reliability was expected to be 100%. These variables included (a) three measures of time-dependent spatial variability (i.e., Slp1, Slp2, and Slp_d), which is a proxy for the fatigue of the articulators; (b) six measures of time-independent spatial variability (i.e., Sse1, Sse2, Sse, Scanning_d1, Scanning_d2, and Scanning_d), which assess cross-cycle or cycle-to-cycle spatial variability; (c) three measures of displacement (i.e., D_max_open, D_max_close, and D_mean), which correspond to the maximum opening, maximum closing, and mean displacement across all cycles; (d) three temporal measures including the cross-cycle temporal variability (i.e., Tsd), the cycle-to-cycle temporal variability (i.e., jitter), and the mean cycle duration (i.e., Tmean); (e) four spatiotemporal measures including the maximum velocity (i.e., max_vel), the spatiotemporal index (STI), which indexes spatiotemporal variability across cycles, the spatiotemporal regularity derived using dynamic time warping (D_dtw), and the mean correlation coefficient between each DDK cycle and a cosine wave (R_mean); and (f) three additional measures of the overall DDK performance including the total duration of the DDK sequence (i.e., Duration), the cycle frequency (F), and the total number of cycles (Num_Cycles). In addition, an alternating motion rate was calculated by dividing the total number of cycles produced by the total sequence duration (DDK rate), yielding a total of 23 variables. These overall measures were included because they are the objective measures of DDK performance typically used clinically. Although these measures are typically derived from the acoustic signal, in this study, they were derived from the kinematic signal as part of the automated MATLAB algorithm.

Statistical Analysis

Because of the large number of variables and small sample size, a descriptive analysis was conducted first to visually examine differences between groups on each of the kinematic variables and used to narrow our statistical analyses to a reduced set of variables. We also wanted to statistically examine group differences in intelligibility to ensure intelligibility was equivalent between groups. A series of Mann–Whitney *U* tests were used to compare groups on the reduced set of kinematic variables derived from the automated DDK analysis that descriptively differed between groups and speech intelligibility. Because this study was exploratory in nature and our objective was to identify quantitative measures that show promise for future investigation, we employed an alpha level of $p < .05$ for all comparisons and included effect sizes (r) to estimate the size of the differences between groups.

Receiver operating characteristic (ROC) analyses were conducted in R (R Core Team, 2017) to compare how well the statistically significant kinematic variables with the largest effect sizes differentiated between children with and without CP, using packages ROCR (Sing et al., 2005), pROC (Robin et al., 2011), and OptimalCutpoints (López-Ratón et al., 2014). ROC analyses yielded a “threshold” for each kinematic variable, the value at which the variable separated children with CP from the TD group with the highest degree of accuracy. This threshold value was then used to calculate the sensitivity (i.e., the measure’s ability to designate children with CP as having CP), specificity (i.e., the measure’s ability to designate children with TD as not having CP), and accuracy of each measure. Area under the curve (AUC) values were calculated to assess the overall strength of each variable for identifying children with CP. Positive predictive value, the probability that children classified as having CP by a measure truly have CP, was also calculated for each kinematic variable.

Results

Group Comparisons

Boxplots were constructed for each of the 23 kinematic variables to visually examine differences between the TD and CP groups. On the basis of this visual analysis of the data, 15 variables had similar means and distributions for both groups and were excluded from further analysis. The remaining seven variables (i.e., duration, D_mean, Tsd, Num_Cycles, STI, slp1, and d_max_open) were included in subsequent statistical analyses.

Results of Mann–Whitney *U* tests on these seven variables are reported in Table 2. Results showed that five of the automated DDK kinematic variables significantly differed between the CP and typically developing groups: Tsd (i.e., the standard deviation of cycle duration) and STI (i.e., STI) were larger in the CP group than the TD group ($p = .04$ and $p = .05$, respectively). D_mean (i.e., the

Table 2. Results of Mann–Whitney *U* tests on kinematic variables passing initial descriptive screening and intelligibility.

Variable	*U*	*Z*	*p*	*r*
Duration	9.00	–2.42	**.02**	.60
D_mean	12.00	–2.10	**.04**	.53
Tsd	12.00	–2.10	**.04**	.53
Num_Cycles	12.50	–2.06	**.04**	.51
STI	13.00	–2.00	**.05**	.50
slp1	14.00	–1.89	.06	.47
d_max_open	16.00	–1.68	.09	.42
Intelligibility	23.50	–0.90	.38	.23

Note. Values in bold indicate a statistically significant difference between groups. STI = spatiotemporal index.

mean displacement of cycles), duration (i.e., duration of the entire DDK sequence), and Num Cycles (i.e., the total number of DDK cycles produced on one breath) were reduced in the CP group compared to the TD group (p = .04, .02 and .04, respectively). Effect sizes were large ($r > .5$) for all statistically significant variables. Intelligibility did not significantly differ between groups. Group differences and individual data for the five significant variables and intelligibility are shown in Figure 1.

ROC Analysis

Results of ROC analyses are shown in Figure 2, and diagnostic accuracy statistics are presented in Table 3. D_mean and duration had the highest diagnostic accuracy for identifying children with CP compared with children with TD. Both variables had 88% sensitivity and specificity. AUC value was highest for duration of the DDK sequence, indicating this variable was the best measure for differentiating groups overall. Positive predictive values (PPVs) indicated that children with DDK sequence durations under 5.5 s had an 88% probability of having CP; however, the confidence interval was large, ranging from 52% to 98% probability.

Figure 1. Boxplots showing the distribution of values in the cerebral palsy (CP) and typical development (TD) groups for the five kinematic variables that differed significantly between groups and the distribution of intelligibility scores. Individual data for all children are displayed as dots superimposed on the boxplots. Red dots indicate children with dysarthria. STI = spatiotemporal index.

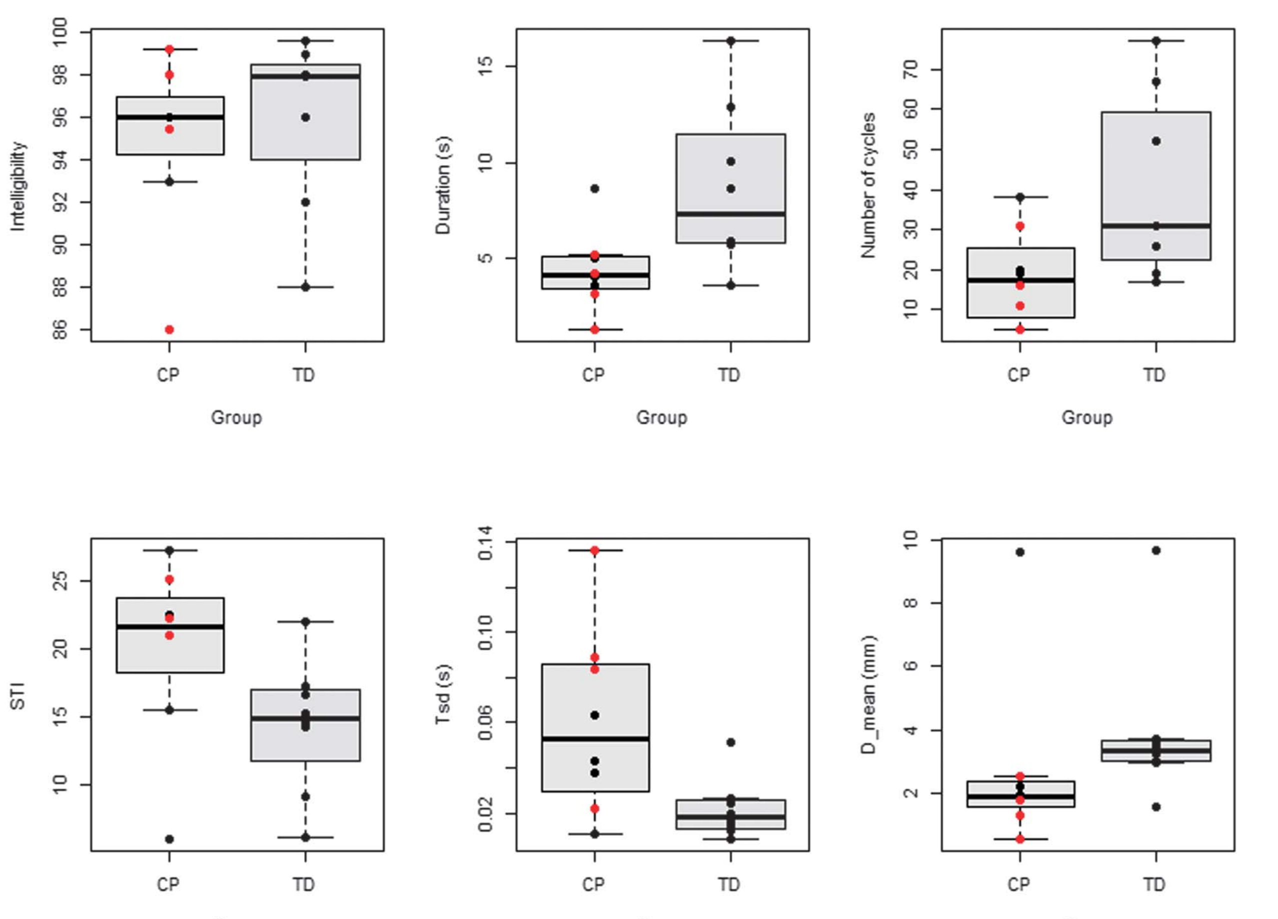
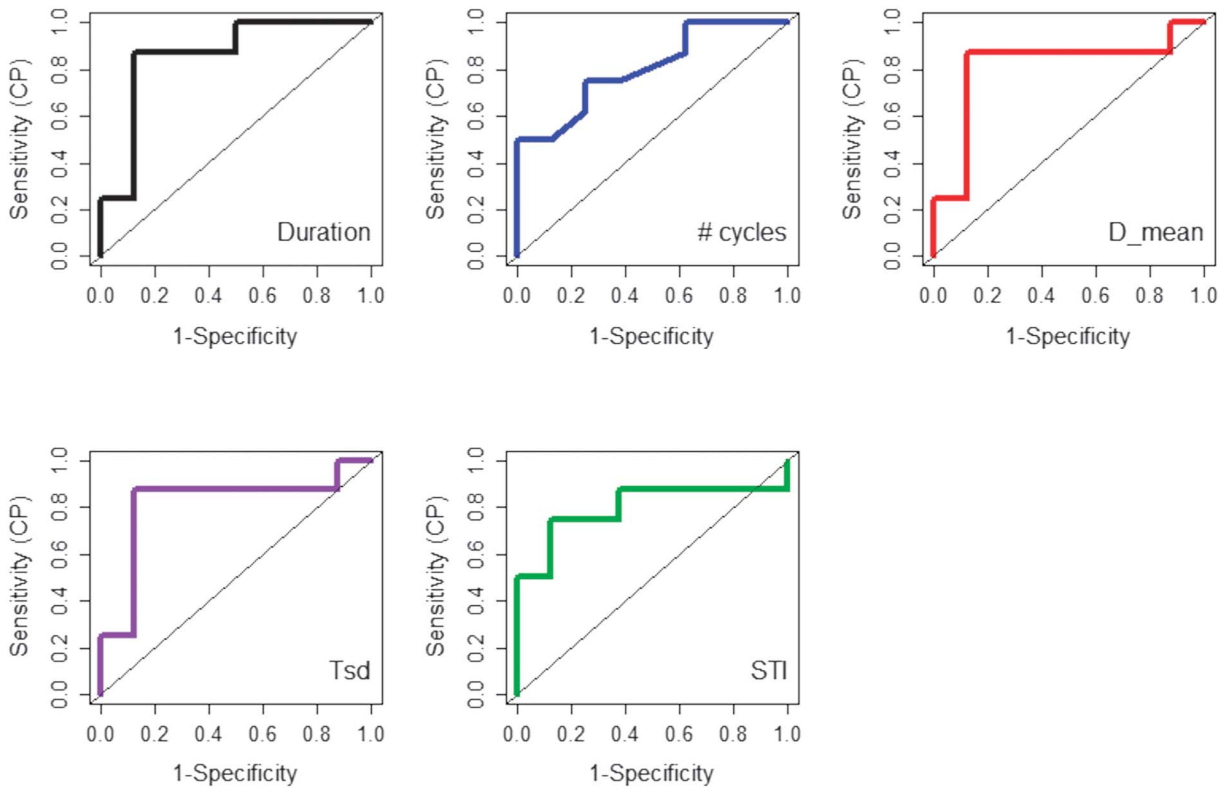


Figure 2. Results of receiver operating characteristic (ROC) analysis. ROC curves are displayed for the five kinematic variables that differed significantly between cerebral palsy (CP) and typical development (TD) groups in separate panels. STI = spatiotemporal index.



Misclassifications of children in the CP group (i.e., children with CP who were classified as TD based on the ROC-derived threshold) were examined for each variable and compared with the child's dysarthria diagnosis. D_mean misclassified one of the four children without a dysarthria diagnosis (CP3). Duration and Num Cycles misclassified a different child without a dysarthria diagnosis (CP6). STI misclassified a third child without dysarthria (CP8) and the same child misclassified by the D_mean variable. Tsd misclassified one of the four children with dysarthria (CP7).

Only one child in the TD group (TD3) was misclassified based on the ROC-derived thresholds. This child was misclassified as belonging in the CP group by all five variables. Notably, this child's sentence intelligibility was 92%, the lowest among the TD group and lower than expected for a 9-year-old. No other remarkable auditory-perceptual features or articulation errors were observed in this child's speech.

Table 3. Diagnostic accuracy statistics for the five kinematic variables that significantly differed between cerebral palsy (CP) and typical development (TD) groups.

Variable	AUC	Threshold	Sensitivity	Specificity	Accuracy	PPV (CI)
Duration (s)	0.86	5.50	0.88	0.88	0.88	88% [52%, 98%]
D_mean (mm)	0.81	2.77	0.88	0.88	0.88	88% [52%, 98%]
Tsd (s)	0.81	0.03	0.88	0.75	0.81	75% [46%, 91%]
STI	0.80	19.14	0.87	0.75	0.81	75% [46%, 91%]
Num_Cycles	0.80	23.00	0.75	0.75	0.75	75% [46%, 91%]

Note. AUC = area under the curve; PPV = positive predictive value; CI = confidence interval.

Discussion

The goal of this study was to explore the ability of automated kinematic measurements of articulatory movement during a DDK task to detect differences in speech motor performance between children with CP who have high intelligibility and children with TD. Results of this study identified five automated kinematic measures of DDK performance that significantly differentiated children with CP from children with typical speech development, despite the groups having equivalent intelligibility levels. Three of the measures, D_mean, Tsd, and STI, reflect different spatial and temporal dimensions of DDK production. The other two measures, DDK sequence duration and number of cycles, relate to the overall quantity of speech output during the task. Duration of DDK sequences and D_mean showed good sensitivity and specificity in this preliminary study for detecting mild speech motor involvement in children. These findings are discussed in detail below.

Spatiotemporal Measures of DDK

Three kinematic variables related to spatial and temporal dimensions of children's DDK cycles showed significant differences between groups. Children with CP had significantly smaller D_mean values, and significantly larger Tsd and STI values compared with children with TD. D_mean is a measure of spatial excursion in lip movements across the DDK sequence. Healthy adult speakers tend to decrease the average displacement of the lips throughout the DDK sequence (Rong et al., 2018). In this study, the opposite effect was demonstrated in children; D_mean was significantly larger for children with typical speech development compared with children with CP. Only two of the eight children in the TD group showed a reduction in lip excursion throughout the sequence, contrasting with prior findings in healthy adults. Because adaptation is a learned motor strategy, it may not be as well adopted in children with an immature oromotor system as in adults, possibly explaining the lack of adaptation in most of the TD children. In the CP group, four of the eight children showed a reduction in lip excursion throughout the sequence. This could be due to task adaptation in the children with CP; however, alternate explanations, such as fatigue or individual differences in motor presentation, should be explored in future research with larger cohorts.

Tsd is the standard deviation of the DDK cycle durations and thus is a measure of temporal variability across the DDK sequence. The larger Tsd values in the CP group indicate greater variability in the duration of syllables produced during the DDK sequence compared with the children with TD. STI, a variable reflecting overall spatiotemporal variability of the DDK cycles, was also significantly larger in the CP group than the TD group, further indicating elevated cycle-to-cycle variability in DDK production among the children with CP. Together, these findings suggest that children with CP have greater spatial and temporal variability in their articulatory movements during DDK production compared to children with TD. Spatial and temporal variability has been shown to be elevated in many pediatric populations, including children with CAS (e.g., Moss & Grigos, 2012) and children with language impairment (e.g., Vuolo & Goffman, 2018); however, it has received minimal study in children with dysarthria. Prior work has shown that spatial and temporal coupling of the upper and lower lips in children with CP, as measured by correlation of kinematic signals, is strongly related to severity as indexed by speech intelligibility (Nip, 2017). Wit et al. (1993) also found greater temporal variability in DDK performance in children with CP and dysarthria compared with TD children. Although coupling differs from variability, underlying deficits in speech motor stability may result in both greater spatiotemporal variability in speech movements and reduced coupling between functionally related articulators. Thus, results of this study contribute additional evidence supporting increased spatiotemporal variability in children with CP, even among those with high intelligibility.

Spatiotemporal measures also showed fairly good sensitivity and specificity for differentiating between children with CP and high intelligibility and children with TD. D_mean showed the strongest sensitivity, specificity, and positive predictive value among the three measures, with 88% accuracy in accurately identifying children with CP. From a clinical perspective, this finding suggests that automated spatiotemporal measures of speech movements may be able to aid in identification of children with speech motor involvement; however, research comparing children with various speech and language disorders is needed before the diagnostic value of these measures can be determined.

Duration of DDK

Results of this study showed that children with CP produced significantly shorter DDK sequence durations and fewer DDK cycles compared to children with TD, despite the fact that the groups did not differ significantly in speech intelligibility. Because DDK is a maximum performance task that requires children to produce syllables quickly and for as long as possible on one breath, the duration and number of cycles produced are influenced by the function of multiple physiologic speech subsystems. Children with CP can have reduced respiratory capacity and/or difficulty with coordination of breath timing for speech due to disrupted neural control of breathing musculature (Solomon & Charron, 1998), both of which could contribute to reduced duration of DDK sequences. Deficits in phonatory valving could also cause inefficient use of breath support and result in decreased duration of the DDK sequence. The number of cycles produced relates to duration (i.e., fewer cycles can be produced in a shorter duration) but is also constrained by articulation rate (i.e., slower articulation rate results in fewer cycles in the same period of time). Prior research has shown that children with CP use larger articulatory excursions during speech compared with children with TD (Nip, 2013; Nip et al., 2017), which may also contribute to a reduction in articulation rate and the number of cycles produced. In this study, DDK rate (number of cycles/second) was descriptively similar between groups and duration of the sequence appeared to be the primary limiting factor in the number of DDK cycles produced.

Furthermore, results of the ROC analysis showed that duration was one of the two most sensitive and specific measures for differentiating children with CP from those with TD, suggesting the potential diagnostic value of this measure. Specifically, having a DDK duration less than 5.5 s identified children with CP with 88% accuracy. Previous research by Thoonen et al. (1996, 1997, 1999) showed that monosyllabic repetition rate during a DDK task and vowel prolongation differentiated children with dysarthria from children with CAS with a high degree of accuracy. In this study, DDK rate did not differ between children with CP and those with TD, which may be due to the high intelligibility and the overall milder speech motor involvement of the included children with CP. The reduced duration of DDK sequences is consistent with Thoonen et al.'s (1996) finding of reduced vowel prolongation in children with dysarthria, both suggesting reduced capacity for sustaining voicing. Although duration and number of cycles were automatically extracted from the kinematic data in this study, these measures can also be easily and reliably extracted from acoustic recordings of the DDK sequence and are commonly used in clinical settings as part of motor speech exams (e.g., Duffy, 2013; Robbins & Klee, 1987). Thus, these findings support the value of including duration measures during maximum performance tasks as part of clinical motor speech exams in children.

Given our small sample size, it is possible the two children with CP who only produced five cycles in the DDK task disproportionately contributed to the sensitivity of the duration measure for differentiating groups. However, even with these two children removed, the mean DDK duration and number of cycles produced was substantially reduced in the CP group (mean duration = 5.1 s; mean number of cycles = 22) compared with the TD group (mean duration = 8.6 s; mean number of cycles = 40). Future studies with larger sample sizes are needed to validate these findings.

These findings also provide information about the speech motor presentations of children with CP who have high intelligibility. In previous research, habitual articulation rate in connected speech has been shown to differentiate children with dysarthria secondary to CP from children with TD (Allison & Hustad, 2018a). Articulation rate has also been shown to be a useful measure for detecting subtle differences in speech motor function even among children with TD (Redford, 2014). Although the children with CP in this study were able to produce syllables at a rapid rate similar to the TD group on the DDK task, it is possible that children's focus on rate resulted in decreased control of airflow and thus reduced duration of the DDK sequences. In other words, children with mild speech motor involvement may have the ability to execute speech tasks with high motor demands, but at the expense of speech dimensions less important to task performance. Therefore, these findings collectively suggest the importance of interpreting the diagnostic strength of speech measures in the context of specific task demands.

Limitations and Future Directions

Results of this study are preliminary and should be interpreted with caution in the context of its limitations. The sample size for this study was small and included a fairly wide age range of children. Thus, results need to be validated with a larger group of participants before findings can be generalized. The misclassification of one child in the TD group suggests the need for establishing robust normative data on these measures before they could be used diagnostically. It is also possible that the children with CP may have shown intelligibility differences from the TD group if they had been asked to produce longer sentences, similar to previous findings (Allison & Hustad, 2014). Future studies should also include additional groups to determine the extent to which automated kinematic measures of DDK can differentiate children with different types of speech motor impairment (e.g., dysarthria vs. CAS) and other speech sound disorders. This study only examined DDK using a "ba" sequence, and it is possible that kinematic measures derived from other DDK tasks (e.g., "pa" or "pataka") may have higher diagnostic accuracy for identifying children with speech motor impairment. In addition, future work relating spatial and temporal measures of DDK production to functional speech measures (e.g., intelligibility and speaking rate) in children with dysarthria may help advance understanding of the biomechanical factors affecting speech production. Differences in spatial and temporal measures between children with different CP subtypes (e.g., spastic vs. dystonic) may also yield valuable insights regarding the influence of different movement disorders on speech production. Data for this project were acquired using two different kinematic systems, which may have introduced additional variability to the data; however, both systems have documented high accuracy in motion tracking and DDK administration protocols and automated kinematic analysis methods were consistent across sites.

Clinical Implications

Findings from this study suggest that DDK sequence duration and number of cycles, which can be easily counted from audio recordings obtained in clinical settings, may be sensitive measures for detecting speech motor involvement in children, thus supporting the value of including DDK as part of clinical motor speech assessment. In addition, the ability of reduced DDK duration to differentiate children with CP from those with TD underscores the importance of considering limitations in respiratory and phonatory subsystems in assessment and treatment with this population. Indeed, three intervention approaches that have mounting evidence for children with CP all involve a focus on respiratory support and/or establishing healthy vocal loudness (i.e., systems approach [Pennington et al., 2010, 2018], Lee Silverman Voice Treatment [Boliek & Fox, 2017], and Speech Intelligibility Treatment [Levy et al., 2021]).

In addition, our findings suggest the possible clinical utility of automated DDK analysis for enhancing speech motor assessment in children. Although kinematic recordings and analysis are not currently used in general clinical settings, efforts are underway to make kinematic systems that are designed for clinical use (e.g., Katz et al., 2014). As these systems become more clinically accessible, development of automated DDK analysis software could lead to more efficient tools for detection of speech motor impairment. Although children with CP who have expected intelligibility for their age may not need speech intervention, objective measures sensitive to speech motor involvement are needed to help resolve diagnostic uncertainties in children who do have reduced speech intelligibility, but the motor speech contribution is unclear. Future work on sensitivity and specificity of these measures may lead to improved identification of children who would benefit from a motor-based intervention approach that leverages principles of motor learning (Maas et al., 2008).

Data Availability Statement

De-identified data are available on request from the authors.

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