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Research Article

Longitudinal Change in Speech Rate and Intelligibility Between 5 and 7 Years in Children With Cerebral Palsy

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Purpose: We examined growth between 5 and 7 years in speech intelligibility, speech rate, and intelligible words per minute (IWPM) in 3 groups of children: those who were typically developing (TD), those with cerebral palsy (CP) and clinical speech motor impairment (SMI), and those with CP and no SMI (NSMI).
Method: Twenty-six children with CP, 16 with SMI, and 10 with NSMI were each seen at 5, 6, and 7 years of age. A cross-sectional group of 30 age-matched TD children, 10 in each age group, were included as controls. All children produced a corpus of utterances of 2–7 words.
Results: All groups of children showed increases in intelligibility and IWPM between 5 and 7 years. Only children with SMI showed increases in speech rate over time. Patterns of change were similar for children in the TD and NSMI groups but different for children in the SMI group.
Conclusions: The window of time between 5 and 7 years is an important period of growth for the production of connected speech where nearly all children, regardless of group, made significant changes in speech intelligibility and IWPM. Interventions focusing specifically on enhancing intelligibility in this age range may help facilitate even further growth in children with SMI, who still had marked intelligibility reductions at 7 years of age.

Cerebral palsy (CP) is the leading cause of motor impairment in children (Reddihough & Collins, 2003) and is associated with disturbances in early brain development before, during, or after birth, resulting in movement and posture difficulties (Bax, Goldstein, Rosenbaum, & Levinton, 2005). Individuals with CP are at risk for a variety of problems related to speech and language, and at least 60% of those with CP have some sort of communication impairment (Bax, Tydeman, & Flodmark, 2006). For many children with CP, neurological involvement affects motor control for speech. Studies suggest that about half of children with CP have dysarthria (Nordberg, Miniscalco, Lohmander, & Himmelmann, 2013), which results from disruption in the performance of any of one or more of the speech subsystems and their associated structures and functions (articulation, phonation, resonance, and respiration; Yorkston, Beukelman, Strand, & Hakel, 2010). Reduced intelligibility is a hallmark feature of dysarthria (Darley, Aronson, & Brown, 1969a); reduced speech rate is also common and is a global indicator of speech motor coordination difficulties across speech subsystems (Yorkston et al., 2010). Previous research has shown that speech rate is an important contributing factor for differentiating between those children with CP who have dysarthria and those who do not (Allison & Hustad, 2018a; Hustad, Gorton, & Lee, 2010).

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Functional Speech Development in Typically Developing Children

Increases in speech intelligibility and in speech rate are important features of speech development in children. In all children, acquisition of intelligible speech is a developmental process for which empirical benchmarks have not been well established, in part because intelligibility is a complex construct that can be defined and measured in a variety of ways. As a result, there is some ambiguity about age expectations for crossing intelligibility thresholds. The few existing studies have important methodological differences that make piecing findings together across studies a difficult endeavor. One methodological problem among

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extant studies is the use of listeners who are "experts," commonly speech-language pathologists (SLPs) or phoneticians/transcriptionists (Austin & Shriberg, 1997; Rice et al., 2010), graduate students in speech-language pathology (Gordon-Brannan & Hodson, 2000), or parent estimates/ratings (Coplan & Gleason, 1988; McLeod, Harrison, & McCormack, 2012). Recent research has demonstrated that there are important differences between experienced listeners and naive listeners (Baudonck, Buekers, Gillebert, & VanLierde, 2009) and that learning occurs for listeners over time (Hustad, Oakes, & Allison, 2015; Liss, Spitzer, Caviness, & Adler, 2002; Tjaden & Liss, 1995). One of the very limited number of studies examining intelligibility development in typically developing (TD) children using naive listeners found that sentence intelligibility was 86% for 3-year-old children, 90% for 4-year-old children, 92% for 5-year-old children, and 97% for 6-year-old children (Hodge & Gotzke, 2014a). There was considerable variability among children within each age band, but variability tended to be reduced as children got older. One limitation was that only 12 children were included in each age band. Additional studies that establish normative expectations for intelligibility development in TD children are needed so that deviations from typical expectations can be identified and quantified in children who may be at risk. Given that improving intelligibility is often a key focus in intervention for individuals with dysarthria, normative information on TD children would provide critical age-based benchmarks. One goal of this study was to quantify change in intelligibility with age in TD children and to examine typical developmental change compared to change in children with CP to begin to understand similarities and differences in growth between groups of children.

As with intelligibility, speech rate also increases with age in children. Previous studies have suggested that rate does not become adultlike in TD children until about 13 years of age (Walsh & Smith, 2002) and that increases in cognitive and linguistic abilities as well as advancing speech motor control abilities may play key roles in accounting for rate changes with age (Nip & Green, 2013). Several studies of TD children have found that articulation rate change may plateau or even decrease in the course of reaching an adultlike level (Hall, Amir, & Yairi, 1999; Walker & Archibald, 2006). However, less is known about development of speech rate, in which durational measures are inclusive of pauses. One study by Hodge and Gotzke (2014a) found that speech rate made slow linear increases between the ages of 3 and 6 years, with considerable variability among children that seemed to decrease with age. At 6 years of age, it is noteworthy, however, that children in the Hodge and Gotzke study had mean speech rates that were considerably below those observed for adults. Development of an adultlike speech rate appears to be a protracted process that emerges gradually over a relatively long time. This study sought to examine change in speech rate among groups of TD children between the ages of 5 and 7 years and to consider the extent to which rate and intelligibility changes covaried over time. We chose this age range because little data exist for children at 7 years of age and this is an important time when speech production is undergoing important refinements. We were also interested in quantifying the extent of change at sequential ages prior to and through this point. Such information will begin to lay the foundation for developing a set of critical benchmarks that enable us to quantify the extent to which a child with CP may be similar to or different from typical expectations on rate and intelligibility measures.

Functional Speech Development in Children With CP

In children with dysarthria, persistent intelligibility problems are common, and although developmental improvements are expected, the rates and limits of change for intelligibility in children with CP have not been established. Children with CP are a heterogeneous population; thus, characterizing intelligibility development necessarily requires the use of longitudinal designs where each child can serve as his or her own control. Studies of this nature are currently underway, and findings suggest that, even by 8 years of age, most children with CP do not surpass 75% intelligibility on single-word productions (Hustad, Sakash, Natzke, Broman, & Rathouz, 2019). Furthermore, recent work in which children with CP are examined from 2 to 8 years of age indicates not only that children show the most growth in single-word intelligibility between 3 and 5 years of age but also that growth continues through 8 years of age (Hustad, Sakash, Natzke, et al., 2019). It is important to note that, in the aforementioned longitudinal study, children with CP were not separated into profile groups based on whether they had speech motor involvement (dysarthria) but, rather, they were considered as a single population. Our work has shown that children with CP differ in important ways based on whether they have speech motor involvement. For example, intelligibility delays and/or deficits are common in children with CP, even in children who do not present with speech motor impairment (SMI; dysarthria). The delays in children without dysarthria, however, are much milder than those observed in children with speech motor involvement (Hustad, Sakash, Broman, & Rathouz, 2019; Hustad, Schueler, Schultz, & DuHadway, 2012). Thus, there are compelling reasons to examine intelligibility development by profile group. In addition, it is clear that children with CP continue to show changes in their speech development through 8 years of age; however, it is not known whether there are differences in growth trajectories due to the presence of SMI.

Most studies to date have looked at preschool-age children with CP. In this study, we focus on children between the ages of 5 and 7 years because little is known about children with CP in this age range and because this is an important time when children are moving through early elementary school. Intelligible speech is critical to school success. Reduced speech intelligibility often disrupts participation in social contexts and can be a barrier to functional independence (Dickinson et al., 2007). Children with CP who have reduced intelligibility, regardless of whether they have SMI, are at a risk for social isolation and difficulties in the classroom. As a result, improving speech intelligibility is often a primary goal of intervention (Hodge & Gotzke, 2014a).

Modifications to speech rate, most commonly reducing an already slow rate, are often targeted as a means to improve speech intelligibility (Pennington, Roelant, et al., 2013). It is unclear, however, how speech rate and intelligibility covary with each other during development and how these functional speech measures are affected by the presence of SMI. Specifically, little is known about how speech rate changes over time in children with CP who have SMI and how it changes over time in children with CP who do not have SMI. In addition, we have limited information regarding how speech rate contributes to overall intelligibility for these children (Lee, Hustad, & Weismer, 2014). In a recent study examining longitudinal change in speech rate, articulation rate, and pausing behavior for children with CP between 4 and 6 years of age, results showed that rate did not change significantly in children with CP, regardless of whether they had SMI (Darling-White, Sakash, & Hustad, 2018). However, Darling-White et al. did find that length of utterance affected speech rate such that longer utterances tended to be produced at a faster rate of speech. The current study expands on the work of Darling-White et al. by examining speech rate and intelligibility in a different set of longitudinal speech samples from older children with CP. We also include a measure of speech efficiency, intelligible words per minute (IWPM; Yorkston & Beukelman, 1981), which simultaneously provides information about speech rate and intelligibility. This hybrid measure has been used in previous studies of children with CP and may provide a more sensitive measure of severity than either intelligibility or rate alone (Hodge & Gotzke, 2014a, 2014b).

In this study, our goal was to examine longitudinal change in speech production among older children with CP during an age when research suggests they should be showing change in their intelligibility (Hustad, Sakash, Natzke, et al., 2019). In particular, we were interested in examining patterns of change in children with CP who had SMI relative to children with CP who did not have SMI. We also examined both groups relative to TD children of the same ages. The primary measures of interest were intelligibility of connected speech, speech rate, and IWPM. This study addressed the following research questions:

1. How do intelligibility, speech rate, and IWPM change from 5 to 7 years of age in children with CP who have SMI, children with CP who have no SMI (NSMI), and TD children?
2. Do the three groups of children show similar patterns of change over time on the three speech production measures?

We hypothesized that all groups of children would show increases in intelligibility and, by association, increases in IWPM (a measure derived from both intelligibility and speech rate). Based on the results of Darling-White et al. (2018), however, we did not expect to see change in speech rate for the groups of children with CP. Finally, we expected that children would show similar patterns of change over time, regardless of group membership, but groups would differ in the magnitude of developmental changes.

Method

Participants

Children With CP

Twenty-six children with CP participated in the present project. All participants were part of a larger longitudinal study on speech and language development in children with CP. To be eligible for participation in the larger study, children were required to have a medical diagnosis of CP and have hearing within normal limits according to distortion product otoacoustic emission screening. Children were recruited for the larger study through neurology and physiatry clinics in the upper midwest of the United States.

For the present project, children were required to have completed at least three data collection sessions between the ages of 5;0 and 7;11 (years;months) in which they produced at least two-word sentences. One of the sessions must have been between 5;0 and 5;11 (Time 1); one, between 6;0 and 6;11 (Time 2); and one, between 7;0 and 7;11 (Time 3). The average age of the children at the 5-year-old visit was 61.9 months (*SD* = 1.6), the average age of the children at the 6-year-old visit was 74.6 months (*SD* = 2.1), and the average age of the children at the 7-year-old visit was 86.1 months (*SD* = 1.6). The 26 children with CP contributed 78[1] total visits. Fourteen of the children were boys; 12 were girls.

Among the 26 children, 10 (seven boys, three girls) had NSMI and 16 (seven boys, nine girls) had SMI. The presence or absence of SMI was determined at each participant's 5-year-old visit by an SLP with expertise in CP. The primary means of diagnosis was through perceptual assessment of each child's speech as produced during the Test of Children's Speech (TOCS+; Hodge & Daniels, 2007). Perceptual assessment of dysarthric speech to identify prominent perceptual features is commonly conducted on standardized speech samples such as these (Darley, Aronson, & Brown, 1969b). Children were also examined for evidence of drooling, facial asymmetry at rest and during movement, and the presence or absence of abnormal tone in the orofacial musculature observed during parent–child interactions and during clinician–child interactions, which were video-recorded for each visit. The second and third authors, who are certified SLPs, classified the children's clinical presentation as SMI or NSMI and also assigned Viking Speech Scale (Pennington, Virella, et al., 2013) ratings to each child.

[1]Note that data from eight of these visits were included in Darling-White et al. (2018).

Classification agreement was 100% for clinical presentation of speech motor involvement; it was also 100% for the Viking Speech Scale. Demographic characteristics of children with CP are provided in Table 1. In addition, at the time of each visit, parents provided information regarding whether their child was receiving speech therapy. These data are presented by child and by profile group in Table 2.

TD Children

Thirty children who were TD (ten 5-year-olds [five boys, five girls], ten 6-year-olds [seven boys, three girls], and ten 7-year-olds [six boys, four girls]) were included as control participants in the current project. Because children who are TD are expected to be more homogeneous than children with CP in their development and because use of cross-sectional data to examine developmental change in children with TD is widely used in the literature, we used a cross-sectional approach to examine age differences in children who are TD. Children in the TD group were recruited from the same local community as children with CP. Participation was solicited through a database of children who attend local schools whose parents had indicated an interest in being contacted for research. Each child completed one visit (30 total visits). The average age of the 5-year-olds was 61.6 months (*SD* = 1.5), the average age of the 6-year-olds was 77.6 months (*SD* = 3.6), and the average age of the 7-year-olds was 87.0 months (*SD* = 3.2). Participants were required to be monolingual speakers of English, have hearing within normal limits as determined by a standard pure-tone hearing screening, and have speech and language skills that were within normal limits as determined by standardized speech and language assessments. The sample comprised 18 boys and 12 girls.

Table 1. Demographic characteristics of participants with cerebral palsy (CP).

Characteristic	NSMI *n* = 10	SMI *n* = 16
Male–female ratio	7:3	7:9
Mean (*SD*) age, months		
Time 1	62.00 (1.79)	61.75 (1.53)
Time 2	74.00 (1.90)	75.13 (2.09)
Time 3	86.45 (1.57)	85.88 (1.50)
Type of CP		
Spastic	10	13
Diplegia	1	4
Hemiplegia (left)	5	4
Hemiplegia (right)	3	2
Triplegia	0	0
Quadriplegia	0	3
Dyskinetic	0	0
Ataxic	0	2
Mixed	0	0
Unknown	1	1
GMFCS at the age of 5 years		
I	8	5
II	2	5
III	0	2
IV	0	4
V	0	0

Note. NSMI = no speech motor impairment; SMI = speech motor impairment; GMFCS = Gross Motor Function Classification System.

Table 2. Children receiving speech and language therapy per parental report by age, individual child, cerebral palsy group, and Viking Speech Scale (VSS) rating.

			Receiving speech therapy		
Child	Group	VSS	Age 5	Age 6	Age 7
1	NSMI	I	1	0	0
2	NSMI	I	0	0	0
3	NSMI	I	0	0	0
4	NSMI	I	0	0	0
5	NSMI	I	0	0	0
6	NSMI	I	0	0	0
7	NSMI	I	0	0	0
8	NSMI	I	0	0	0
9	NSMI	I	1	1	1
10	NSMI	I	0	0	0
11	SMI	II	1	1	0
12	SMI	II	0	0	0
13	SMI	II	1	1	1
14	SMI	III	1	1	1
15	SMI	II	1	0	1
16	SMI	II	1	1	1
17	SMI	II	1	1	1
18	SMI	II	1	1	1
19	SMI	II	1	1	1
20	SMI	II	MD	1	1
21	SMI	II	0	0	1
22	SMI	II	1	1	1
23	SMI	II	1	1	1
24	SMI	II	0	0	0
25	SMI	II	1	1	1
26	SMI	II	1	1	1

Note. NSMI = no speech motor impairment; SMI = speech motor impairment; MD = missing data.

We used the Arizona Articulation Proficiency Scale–Third Edition (Fudala, 2001) to assess segmental speech development in children who are TD. Children were considered to have passed if they scored within one 1 *SD* from the mean. We used the Preschool Language Scale–Fourth Edition (Zimmerman, Steiner, & Pond, 2002) to assess language development for children aged 5 and 6 years and the Test of Narrative Language (Gillam & Pearson, 2004) to assess language development in children who were 7 years of age. For the Preschool Language Scale–Fourth Edition, in accordance with the technical manual, 5-year-old children passed if they received a total language score of 3 or more out of 6; 6-year-old children passed if they received a language total score of 5 or more out of 6. The 7-year-old children passed the Test of Narrative Language if they scored within one 1 *SD* of the test's normative age mean.

Adult Listeners

Two hundred sixteen adult listeners provided intelligibility ratings of children's speech. Listeners were recruited from the University of Wisconsin–Madison community via public postings. Listeners were primarily undergraduate students and were compensated either monetarily or with extra credit in a communication sciences and disorders class. Listeners were required to be between the ages of 18 and 45 years; be a native speaker of American English; have no identified language, learning, or cognitive disabilities per self-report; and pass a standard pure-tone hearing screening. Among the 216 listeners, 54 were male and 162 were female. The mean age of listeners was 21.39 years (*SD* = 3.65).

Materials and Procedure

Acquisition of Speech Samples From Children

All speech samples were collected from children in a sound-attenuating booth during a data collection session with a certified SLP. Speech samples from the TOCS+ (Hodge & Daniels, 2007) were obtained during the children's visits. Each child repeated individual words and sets of utterances ranging from two to seven words in length following a prerecorded auditory model presented on an iPad. There were 10 utterances of each length, and the utterances comprised phrases that were developmentally appropriate for young children in terms of lexical, phonetic, syntactic, and morphological features. To ensure the speech samples did not overlap with the prerecorded adult model and did not contain environmental sounds, a research assistant monitored each production and asked the child to repeat utterances that contained any noises in the background or that did not contain all of the target words. Only the two- to seven-word TOCS+ utterances were used in data analysis for this study. Not all children produced utterances of all lengths due to speech motor constraints. Table 3 shows the number of children, by group, who produced utterances of each length at each age point.

All speech samples were recorded with professional-quality digital audio and video equipment. Each child repeated the utterances into an Audio-Technica AT4040 condenser studio microphone, which sat 18 in. away from the child. Using a Mackie 12010 VLZ mixer, a research assistant monitored the level of the auditory signal as the child was speaking.

Acquisition of Intelligibility Data

Research assistants trained in acoustic analysis segmented and peak amplitude normalized the speech stimuli in the recording and editing software program Audacity Version 2.1.1. Samples were normalized to ensure that maximum loudness levels of the recorded speech samples were the same across children and utterances, while preserving the amplitude contours of the original productions. This also enabled calibration to peak output levels for playback to listeners. All individual utterances were compiled into a randomized control file to be played for listeners.

Adult listeners were seated individually in a sound-attenuated booth in front of a keyboard, a 19-in. flat-screen computer monitor, and an external speaker. The external speaker was calibrated regularly by a research assistant to ensure the peak output level was 75 dB SPL from where listeners were seated.

Listeners were told that the purpose of the study was to determine how understandable children's speech is by unfamiliar adult listeners like themselves. Speech stimuli were delivered in random order to each listener via the external speaker using an automated computer program. After each utterance was played, listeners orthographically transcribed what they heard. Listeners heard each utterance one time and were instructed to take their best guess if they could not fully understand what the child said. Listeners practiced

Table 3. Number of children who produced each utterance length by group and age.

Utterance length	NSMI (*n* = 10)	SMI (*n* = 16)	TD (*n* = 10 per age group)
Two words	Age 5: 10	Age 5: 16	Age 5: 10
(20 words across all)	Age 6: 10	Age 6: 16	Age 6: 10
	Age 7: 10	Age 7: 16	Age 7: 10
Three words	Age 5: 10	Age 5: 14	Age 5: 10
(30 words across all)	Age 6: 10	Age 6: 16	Age 6: 10
	Age 7: 10	Age 7: 16	Age 7: 10
Four words	Age 5: 10	Age 5: 13	Age 5: 10
(40 words across all)	Age 6: 10	Age 6: 15	Age 6: 10
	Age 7: 10	Age 7: 16	Age 7: 10
Five words	Age 5: 10	Age 5: 13	Age 5: 10
(50 words across all)	Age 6: 10	Age 6: 15	Age 6: 10
	Age 7: 10	Age 7: 15	Age 7: 10
Six words	Age 5: 10	Age 5: 12	Age 5: 10
(60 words across all)	Age 6: 10	Age 6: 14	Age 6: 10
	Age 7: 10	Age 7: 15	Age 7: 10
Seven words	Age 5: 9	Age 5: 10	Age 5: 8
(70 words across all)	Age 6: 10	Age 6: 14	Age 6: 10
	Age 7: 10	Age 7: 15	Age 7: 10

Note. NSMI = no speech motor impairment; SMI = speech motor impairment; TD = typically developing.

two sample items with a research assistant to become oriented with the software and the task.

Two listeners made orthographic transcriptions of each child at each age point to prevent potential learning effects associated with hearing the same speakers over time and/or the same speech stimuli produced repeatedly. Each word transcribed by listeners was scored as either correct or incorrect relative to the target utterance that the speaker attempted. Homonyms and misspellings were accepted as correct if the phonemes from the spoken version of the utterance matched what the listener typed. The total number of words identified correctly across the two listeners for all utterances was divided by the total number of words possible (across the two listeners) and multiplied by 100 to yield a percent intelligibility score for each child and each visit. In other words, the intelligibility score measured the overall percentage of words successfully recovered by two unfamiliar adult listeners, and we computed one of these scores for each visit by each child.

Acquisition of Speech Rate Data

Research assistants trained in acoustic analysis used the software program Praat (Boersma & Weenink, 2015) to mark the initiation and termination of each segmented and normalized two- to seven-word utterance. To do so, research assistants created text grids to align the spectrogram, waveform, phonemes, and words into one visible display. The research assistants then marked the initiation and termination of each utterance. The initiation of the utterance was identified by the onset of audible or visible acoustic energy at the onset of the first phoneme produced by the child, not including audible inspiration or environmental noise. The termination of the utterance was identified by the offset of acoustic energy at the offset of the final phoneme in the utterance. Speech rate, in words per minute (wpm), was calculated by adding the total number of words a child spoke across all utterances, divided by the total duration of all utterances in seconds (inclusive of all pauses) and multiplied by 60 s.

Acquisition of IWPM Data

IWPM was calculated in the same way as speech rate but used the number of intelligible words instead of the words spoken by the child. Specifically, we added the mean (across the two listeners) of the total number of words identified correctly for each child, divided by the total duration of all utterances in seconds, and multiplied by 60 s.

Experimental Design and Statistical Procedure

Research questions of interest focused on examining (a) statistical differences over time within groups for each of the three measures, namely, intelligibility, speech rate, and IWPM, and (b) a descriptive comparison between groups and measures regarding patterns of change over time. A 1 × 3 design was employed to examine differences over age points (5, 6, and 7 years) within each of the three groups of children (NSMI, SMI, and TD) on each of the three dependent measures (intelligibility, speech rate, and IWPM). For the two groups of children with CP (NSMI and SMI), data were longitudinal in nature; thus, one-way repeated-measures analyses of variance (ANOVAs) were performed along with dependent-samples follow-up contrasts. For children in the TD group, data were cross-sectional in nature; thus, between-subjects ANOVAs were performed along with independent-samples follow-up contrasts. Use of mixed methods (cross-sectional and longitudinal) is widely used in developmental research in which growth is a key variable of interest. See, for example, articles from the World Health Organization describing growth curves for height and weight (de Onis et al., 2006, 2007; WHO Multicentre Growth Reference Study Group, 2006). For each omnibus test, a p value of .01 or less was required for significance (total error rate = .03 per family of omnibus tests, where a family is defined as tests conducted within groups of children). A p value of .01 or less was also required for significance for each follow-up test (total potential error rate of .09 per family of tests, where a family is defined as tests conducted within groups of children).

Results

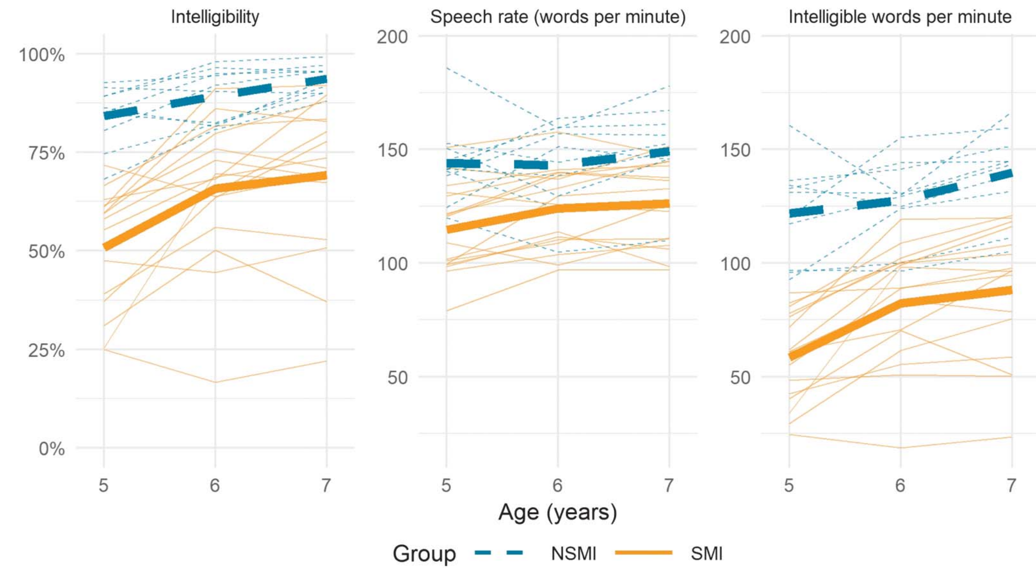
Intelligibility Differences Over Time Within Groups

Figure 1 shows longitudinal intelligibility change over time for the two groups of children with CP; Figure 2 shows intelligibility change by age for cross-sectional groups of children with TD. Table 4 provides descriptive data for all children in tabular form. Descriptive results for group data suggest that there was steady improvement in speech intelligibility over time for each of the three groups; however, there was considerable variability among children within each group, particularly for children with SMI. Inferential statistics, provided in Table 5, revealed significant omnibus ANOVA results for each of the three groups. Pairwise tests between time points within groups, shown in Table 6, revealed that differences between 5 versus 7 years of age were significant for all three groups. However, the difference between 5 versus 6 years was significant only for the SMI group, and the difference between intelligibility scores at 6 versus 7 years was significant only for the NSMI group.

Speech Rate Differences Over Time Within Groups

Figure 1 also shows longitudinal speech rate change over time for the two groups of children with CP; Figure 2 shows speech rate change by age for cross-sectional groups of children with TD. Descriptive results suggest that there was a slight increase in speech rate over time for children in the SMI group but that performance of children in the NSMI group and the TD group did not show much change. Considerable variability for individual children within groups was again present. Inferential statistics, provided in Table 5, revealed nonsignificant omnibus ANOVA

Figure 1. Longitudinal change in intelligibility (as measured by percentage of words identified correctly), speech rate, and intelligible words per minute for children with cerebral palsy (CP) at 5, 6, and 7 years of age. Note that thick lines represent means by profile group, and thin lines represent individual child data. The no speech motor impairment (NSMI) group comprises children with CP and no (clinical) speech motor impairment; the speech motor impairment (SMI) group comprises children with CP and (clinical) speech motor impairment.



results for children in the TD group and children in the NSMI group. However, the omnibus ANOVA was significant for children in the SMI group. Pairwise contrasts between time points for the SMI group, shown in Table 6, revealed that differences between 5 versus 6 years and between 5 versus 7 years were statistically significant.

IWPM Differences Over Time Within Groups

Figure 1 also shows longitudinal change in IWPM over time for the two groups of children with CP, and Figure 2 shows IWPM change by age for cross-sectional groups of TD children. Descriptive results suggest that there was steady improvement in IWPM over time for each of the

Figure 2. Cross-sectional differences by age for intelligibility (as measured by percentage of words identified correctly), speech rate, and intelligible words per minute for typically developing children at 5, 6, and 7 years of age. Note that thick lines represent averages (with error bars representing standard deviations), and dots represent individual child data.

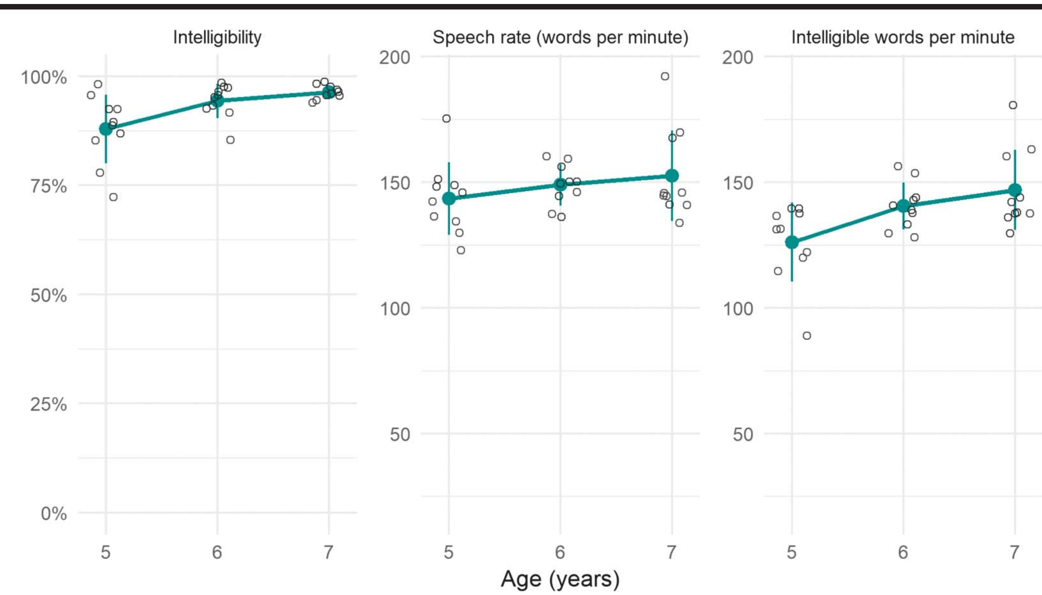


Table 4. Descriptive results for intelligibility, intelligible words per minute, and speech rate by group and age.

Measure and age	NSMI (n = 10)	SMI (n = 16)	TD (n = 10 per year)[a]
Intelligibility ($M \pm SD$)			
Age 5	84 ± 7.8	51 ± 14.9	88 ± 7.9
Age 6	89 ± 6.9	66 ± 18.1	94 ± 3.9
Age 7	94 ± 3.6	69 ± 19.6	96 ± 1.5
Intelligible words per minute ($M \pm SD$)			
Age 5	122 ± 21.7	58 ± 20.2	126 ± 15.8
Age 6	128 ± 18.3	82 ± 26.1	141 ± 9.3
Age 7	140 ± 19.4	88 ± 29.3	147 ± 15.9
Speech rate in words per minute ($M \pm SD$)			
Age 5	144 ± 17.9	115 ± 19.6	144 ± 14.4
Age 6	143 ± 19.0	124 ± 18.0	149 ± 8.2
Age 7	149 ± 19.5	126 ± 18.6	153 ± 18.0

Note. NSMI = no speech motor impairment; SMI = speech motor impairment; TD = typically developing.

[a]Children with cerebral palsy participated longitudinally. The typically developing cohort is a cross-sectional sample (different children at each age).

three groups. Inferential statistics, provided in Table 5, revealed significant omnibus ANOVA results for each of the three groups. Pairwise tests between time points within groups, shown in Table 6, revealed that differences between 5 versus 7 years were statistically significant within each group. However, the difference between IWPM at 6 versus 7 years was significant only for the children in the NSMI group, and the difference between 5 versus 6 years was significant only for the SMI group.

Descriptive Patterns of Change Among Variables and Groups

Examination of patterns of change over time within groups suggests that children with TD had a significant 8% increase in intelligibility, beginning at 88% at 5 years and ending at 96% at 7 years. However, the cross-sectional results for TD children showed no change in rate of speech between any of the time points, with mean speech rates of about 150 wpm throughout the age range. IWPM changed by about 20 wpm between the ages of 5 and 7 years, increasing from 126 to 146 IWPM.

Children in the NSMI group were very similar to children in the TD group in their patterns of change. For intelligibility, they showed a nine percentage point increase between 5 and 7 years, beginning at 84% and ending at 93%. Children in the NSMI group also showed a significant increase of 4% in intelligibility between 6 and 7 years of age. Like children in the TD group, those in the NSMI group showed no change in their speech rate, with mean speech rates slightly below 150 wpm throughout the age range. IWPM increased by 18 wpm between the ages of 5 and 7 years, changing from 117 to 135 IWPM. These children also had a significant increase of 12 IWPM between 6 and 7 years of age. Individual data shown in Figure 1 indicate that there was considerable variability among children in the NSMI group. About half of the children showed change in intelligibility with age that was greater than 10 percentage points (up to a maximum of about 20). Although overall similar patterns, including magnitudes of change, were observed in children in the TD and NSMI

Table 5. Analysis of variance omnibus test results for each measure nested within group.

Source	df	Mean square	F	p	η_p^2
TD children					
Intelligibility	2	194.351	7.305	.003	.351*
Error	27	26.604			
Speech rate	2	208.011	1.039	.368	.071
Error	27	200.244			
Intelligible words per minute	2	1125.813	5.759	.008	.299*
Error	27	195.500			
NSMI children					
Intelligibility	1.777	251.371	13.706	< .001	.604*
Error	15.995	18.340			
Speech rate	1.125	195.820	1.017	.348	
Error	10.126	192.595			
Intelligible words per minute	1.249	1365.926	8.345	.010	.481*
Error	11.242	163.675			
SMI children					
Intelligibility	1.599	1923.961	19.371	< .001	.568*
Error	23.984	97.511			
Speech rate	1.850	640.792	11.378	< .001	.431*
Error	27.750	56.317			
Intelligible words per minute	1.463	5412.865	29.354	< .001	.662*
Error	21.950	184.397			

Note. Greenhouse–Geisser corrections are reported for repeated-measures omnibus tests for SMI and NSMI groups. Corrected models are also reported for omnibus tests of between-subjects effects for the TD group. TD = typically developing; NSMI = no speech motor impairment; SMI = speech motor impairment.

*p < .01.

Table 6. Follow-up contrasts for significant analysis of variance results.

Contrast	Mean difference	*t*	*SE*	*p*
TD children				
Intelligibility				
6 vs. 5	6.432	–2.311	2.783	.038
7 vs. 5	8.438	–3.317	2.544	.008*
7 vs. 6	2.006	–1.519	1.321	.155
Intelligible words per minute				
6 vs. 5	14.321	–2.478	5.780	.026
7 vs. 5	20.722	–2.929	7.075	.009*
7 vs. 6	6.400	–1.100	5.817	.289
NSMI children				
Intelligibility				
6 vs. 5	5.07	2.59	1.96	.029
7 vs. 5	9.44	4.82	1.96	.001*
7 vs. 6	4.37	3.01	1.45	.010*
Intelligible words per minute				
6 vs. 5	5.85	0.98	5.98	.354
7 vs. 5	18.09	4.54	3.98	.001*
7 vs. 6	12.24	3.96	3.09	.003*
SMI children				
Intelligibility				
6 vs. 5	15.08	4.35	3.47	.001*
7 vs. 5	18.39	5.24	3.52	< .001*
7 vs. 6	3.13	1.50	2.21	.154
Speech rate				
6 vs. 5	9.38	3.76	2.49	.002*
7 vs. 5	11.41	3.97	2.87	.001*
7 vs. 6	2.02	0.90	2.25	.382
Intelligible words per minute				
6 vs. 5	23.87	5.35	4.46	< .001*
7 vs. 5	29.69	6.09	4.88	< .001*
7 vs. 6	5.82	2.22	2.63	.043

Note. TD = typically developing; NSMI = no speech motor impairment; SMI = speech motor impairment.
$^*p < .01$.

groups across variables, it is noteworthy that those in the NSMI group began and ended with intelligibility thresholds and IWPM thresholds below those of children with TD.

Children with SMI showed slightly different patterns of change than the other two groups of children. For intelligibility, they showed an 18 percentage point increase between 5 and 7 years, beginning at 50% and ending at 68%. They also showed significant gains between 5 and 6 years (15 percentage point increase). Unlike children in the TD and NSMI groups, children in the SMI group showed increases in their speech rate of 11 wpm between 5 and 7 years of age, changing from 113 to 124 wpm. As with intelligibility, these children also showed significant change in speech rate between 5 and 6 years of age, increasing around 9 wpm in this time frame. IWPM changed by 30 wpm between the ages of 5 and 7 years, increasing from 57 to 87 IWPM, and again showed significant change between 5 and 6 years (an increase of 24 IWPM). Individual data, shown in Figure 1, suggest that there was more variability among children with SMI than children with NSMI. Children with SMI showed intelligibility that increased by up to 42 percentage points, with 11 children showing gains of 10 or more. Only three children showed intelligibility gains less than 10, and two showed negative intelligibility change. Patterns of change were similarly variable for IWPM and for speech rate.

Discussion

The purpose of this study was to quantify change in speech intelligibility, speech rate, and IWPM between the ages of 5 and 7 years in three groups of children, namely, those who are TD, those with CP and NSMI, and those with CP who have SMI. We used a prospective longitudinal design to examine age differences in children with CP and a cross-sectional design to examine age differences in children with TD. We also sought to examine whether groups of children showed similar patterns of change over time. Key findings from this study were the following: (a) All groups of children showed change over time in intelligibility and IWPM, and children with SMI showed greater change than those in other groups, but there was considerable variability within all groups of children; (b) only children with SMI showed change in speech rate over time, but again there was considerable individual variability; and (c) children with NSMI and children with TD had very similar patterns of change on all measures; however, children with NSMI had slightly reduced intelligibility and IWPM relative to children with TD. These findings are discussed in detail below.

Change Over Time

A key finding from this study was that all groups of children showed change over time in speech intelligibility and IWPM between 5 and 7 years of age. Children with NSMI and those with TD showed gains of about 10 percentage points and reached 94% and 96% intelligibility at 7 years, respectively. Both groups were very close to the ceiling of 100% intelligibility by 7 years of age. It is noteworthy, however, that both groups of children were under 90% intelligible on average at 5 years of age, indicating that intelligibility had not yet reached an adultlike level. This finding is in contrast to previous clinical guidelines suggesting that intelligibility of children with TD should be 100% by 4 years of age (Coplan & Gleason, 1988; Flipsen, 2006). These discrepant findings are most likely due to different methodologies between studies and highlight the importance of informed clinical decision making regarding which measures are used to quantify intelligibility. Regardless, findings of this study show clear evidence that children are continuing to improve their intelligibility as they approach expected mastery thresholds for speech-sound acquisition.

Of the three groups of children examined in this study, those with SMI showed the greatest change in intelligibility, with nearly a 20-point improvement on average over the 2-year time span of this study. They also showed significant improvement between 5 and 6 years of age as a group—but not between 6 and 7 years of age—suggesting that children were growing more rapidly at earlier ages than at later ages. Other work from our laboratory examining growth in single-word intelligibility across a range of children with CP, including those with NSMI and SMI, has found that children showed the greatest intelligibility growth between the ages of 3 and 5 years (Hustad, Sakash, Natzke, et al., 2019). However, the present findings extend our other work, suggesting that the narrower subset of children with CP who have SMI continue to have considerable growth in intelligibility up to 7 years of age, and perhaps beyond.

In this study, children with SMI had considerable variability in their intelligibility and in the magnitude of change in intelligibly over time. Individual data indicate that intelligibility scores for children with SMI spanned from a low of about 20% at 5 years to a high of about 70% at 5 years, clearly reflecting a range of severity levels within the participant pool. Data further indicate that all but two children with SMI showed improvement between 5 and 7 years of age. The magnitude of change varied considerably, however, ranging from a low of a 3-point improvement to a high of 42-point improvement. There did not seem to be a discernable pattern regarding which children had the greatest change. In fact, of the four children with the lowest intelligibility scores at the age of 5 years, two children had very little or negative change, and two children had change in intelligibility scores in the 40% range.

One potential explanation for variability in the SMI group might be the impact of speech therapy. However, we examined cursory data obtained at the time of each visit regarding which children were receiving speech therapy (see Table 3). Findings, based on parent report of speech treatment, do not seem to suggest a clear relationship between change in speech intelligibility and whether or not children were receiving therapy. It is critical to note, however, that we did not have information describing the specific nature of speech therapy, the frequency of therapy, and the duration of therapy that children were receiving. All of these are important variables that may impact change associated with intervention. Further research is needed to investigate these complex relationships.

Another important finding for the SMI group was that intelligibility was considerably reduced at each time point compared to children with TD and NSMI. While children with SMI made progress in their speech intelligibility over time, they still lagged far behind age expectations, and most children had significant functional speech deficits, which we define as intelligibility scores below 80% based on our experience with this population.

In addition to improvements in intelligibility, children with SMI also showed an increased speech rate between 5 and 7 years. Concurrent gains in speech rate and intelligibility may be due to refinements in speech motor control, and particularly coordination, associated with development along with continued speech-sound acquisition and refinement in children with SMI. Previous research on speech development has suggested that many children with CP are delayed in the onset of their ability to speak (Hustad, Allison, McFadd, & Riehle, 2014; Hustad et al., 2017). This later onset of speech may result in a time-shifted period of speech motor development that occurs at slightly older ages for these children, potentially accounting for increases in speech rate observed up to 7 years of age in this study for children with SMI. It is interesting to note that our findings for children with SMI were different than those reported by Darling-White et al. who found no change in speech rate over time. One explanation may be that Darling-White et al. included only children who could produce utterances of four words or longer, while we included children who could produce utterances of two words or longer. This difference may have resulted in a wider range of severity levels in this study, which may account for the difference in speech rate findings.

Findings for children with SMI in this study are in contrast to our results for children with TD and NSMI who did not show changes in their speech rate between 5 and 7 years of age but did show improvements in intelligibility. One explanation is that intelligibility changes for children with TD and NSMI may be associated with refinements in the production of more complex, later acquired speech sounds, known to be reaching mastery levels through 8 years of age (Sander, 1972; Smit, Hand, Freilinger, Bernthal, & Bird, 1990), but not associated with global changes in coordination and speech motor control as reflected by speech rate increases that may have occurred at earlier ages. Findings from this study for children with TD are generally consistent with the very limited previous research examining growth in intelligibility, speech rate, and IWPM in children based on the production of elicited sentences from the TOCS+. However, children with TD in this study were slightly less intelligible at 5 and 6 years of age and had slightly faster speech rates and higher rates of IWPM than children in previous studies that were comparable in nature (Hodge & Gotzke, 2014a). Explanations for these small differences in findings may be attributable to methodological differences between the two studies. For example, listeners in the Hodge and Gotzke study were able to hear sentences twice, which may have resulted in increased intelligibility scores. Children in this study produced more sentences of each word length, which may have inflated speech rate findings given that recent work suggests children may produce longer utterances at a faster rate (Darling-White et al., 2018).

IWPM is a hybrid measure that is dependent on both intelligibility and speech rate. This measure has been used as an index of communication efficiency (Yorkston & Beukelman, 1981), providing enhanced functional information about speech performance beyond individual measures of intelligibility and speech rate. It has been defined as "a combination of the rate and accuracy with which spoken messages are transmitted from speaker to listener" (Yorkston & Beukelman, 1981, p. 297). In this study, findings related to IWPM paralleled those of intelligibility, such that all groups of children showed improvement from 5 to 7 years, but the magnitude of change was greatest for children with SMI.

Patterns of Change Among Groups

Generally speaking, patterns of change over time were very similar for children with NSMI and those with TD. Specifically, both groups of children showed increases in intelligibility and IWPM between 5 and 7 years, but neither group showed changes in speech rate during this time frame. One noteworthy finding was that, although the magnitude of change was similar on each variable for each group of children, children with NSMI generally had lower intelligibility and reduced IWPM than their TD counterparts. Children with NSMI also had greater variability, as a group, than children with TD. These findings are consistent with other studies examining children with NSMI relative to typical expectations (Hustad, Sakash, Broman, et al., 2019) and suggest that children with NSMI may be delayed in their speech development, falling slightly below the performance of typical peers but still above the performance of SMI peers with clear clinical dysarthria (Hustad, Sakash, Broman, et al., 2019; Hustad et al., 2012).

Children with SMI had a different pattern of change from other children in this study. They showed steady improvement on all functional speech measures, with larger changes tending to occur earlier rather than later. The finding of consistent change in speech development for children with SMI through the age of 7 years may have promising implications for treatment. Targeted therapies such as those employed by Pennington and colleagues (Pennington, Lombardo, Steen, & Miller, 2018; Pennington, Miller, Robson, & Steen, 2010; Pennington, Roelant, et al., 2013) that are specific to speech motor profiles, such as those identified by Allison (Allison & Hustad, 2018b), may be particularly beneficial when children are in periods of enhanced growth.

Limitations and Future Directions

In this study, we examined a relatively small group of children with CP over a relatively short amount of time. Similarly, our cross-sectional sample of children with TD was also small, and the age range was narrow to be consistent with longitudinal data from children with CP. Studies that extend the age range of study into adolescence and beyond are necessary to further characterize change in functional speech measures and advance our understanding of the rates and limits of change in speech among children with CP and to determine when children become fully adult-like. In addition, studies that employ larger numbers of children are necessary to ensure that there is sufficient statistical power to find differences.

Studies that establish benchmarks and cut-points for speech intelligibility development and speech rate development for TD children do not currently exist across the range of development. Such studies are necessary to provide normative comparisons against which children with CP, whether NSMI or SMI, can be compared. This kind of data would enable us to quantify how close to or far from typical a child is at any given age and would be useful for intervention decision making.

In this study, we used elicited speech samples so that we could have an accurate frame of reference for the target words children were producing and thus more accurately measure rate of speech and speech intelligibility. Spontaneous speech may look different in terms of speech intelligibility and speech rate and therefore should be examined to further our understanding of how functional speech parameters change with time in both TD children and children with CP.

We did not examine underlying acoustic features of speech that may account for intelligibility and speech rate findings in this study. Studies using a finer grained approach such as acoustic analyses of segmental and suprasegmental features as well as kinematic or physiological measures may provide important explanatory information for the findings observed in this study and may lead to targeted interventions that improve intelligibility and speech rate in children with CP.

Many children in this study were receiving speech/language intervention. However, we were not able to quantify the specific nature, frequency, duration, or goals/objectives targeted in therapy, so we cannot quantify the extent to which intervention may have facilitated or even been the cause of some of the changes seen in children's speech production over time. Future studies should seek to examine this more closely to better understand the potential benefits of therapy in facilitating developmental change in speech production for children with CP.

Children with SMI were heterogeneous in this study. Clinical classifications using the Viking Speech Scale (Pennington, Virella, et al., 2013) revealed that the majority of children with SMI had scores of 2, indicating that the severity of speech involvement was mild to moderate and characterized by imprecision, but was typically understandable to unfamiliar listeners. Despite similar severity categories, there was considerable variability among children with SMI in intelligibility and speech rate. Future studies should seek to characterize sources of variability and to control severity in order to better understand developmental change and its contributors.

Clinical Implications

Despite the limitations of this study, there are several important clinical implications. First, all groups of children are making important changes in speech intelligibility between the ages of 5 and 7 years. For TD children and children with CP and NSMI, intelligibility improvements are on the order of about 10 percentage points. It is noteworthy that not even TD children reached 100% intelligibility in connected speech by 7 years of age, although they did come close. For children with CP and SMI, improvements in intelligibility were slightly under 20 percentage points on average and were thus considerably larger than those for children with TD and children with NSMI. Children with SMI were also making simultaneous changes in their speech rate and were becoming more efficient with their speech, producing more IWPM as they got older. Collectively, these findings suggest that the window of time between 5 and 7 years is an important period of growth for connected speech and that interventions seeking to enhance intelligibility are very likely to play an important role in improving functional communication for children with SMI who, as a group, still have considerable intelligibility deficits at 7 years of age.

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