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Research Article

Longitudinal Growth in Single-Word Intelligibility Among Children With Cerebral Palsy From 24 to 96 Months of Age: Effects of Speech-Language Profile Group Membership on Outcomes

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Purpose: We examined whether there were differences among speech-language profile groups of children with cerebral palsy (CP) in age of crossing 25%, 50%, and 75% intelligibility thresholds; age of greatest intelligibility growth; rate of intelligibility growth; maximum attained intelligibility at 8 years; and how well intelligibility at 36 months predicts intelligibility at 96 months when group membership is accounted for. Profile groups were children with no speech motor impairment (NSMI), those with speech motor impairment and language comprehension that is typically developing (SMI-LCT), and those with speech motor impairment and language comprehension impairment (SMI-LCI).

Method: Sixty-eight children with CP were followed longitudinally between 24 and 96 months of age. A total of 564 time points were examined across children (*M* = 8.3 time points per child, *SD* = 2.6). We fitted a nonlinear random effects model for longitudinal observations, allowing for differences between profile groups. We used the fitted model trajectories to generate descriptive analyses of intelligibility growth by group and to generate simulations to analyze how well 36-month intelligibility data predicted 96-month data accounting for profile groups.

Results: Children with CP who have NSMI have different growth and better intelligibility outcomes than those with speech motor impairment. Children with SMI-LCT tend to have better outcomes but similar intelligibility growth as children with SMI-LCI. There may be a subset of children that cut across SMI-LCI and SMI-LCT groups who have severe speech motor involvement and show limited growth in intelligibility.

Conclusions: Intelligibility outcomes for children with CP are affected by profile group membership. Intelligibility growth tends to be delayed in children with speech motor impairment. Intelligibility at 3 years is highly predictive of later outcomes regardless of profile group. Intervention decision making should include consideration of early intelligibility, and treatment directions should include consideration of augmentative and alternative communication.

Children with cerebral palsy (CP) are at risk for communication challenges, and the majority experience some type of communication disorder (Bax, Tydeman, & Flodmark, 2006; Mei, Reilly, Reddihough, Mensah, & Morgan, 2014; Nordberg, Miniscalco, Lohmander, & Himmelmann, 2013). It is estimated that approximately half of children with CP have dysarthria (Nordberg et al., 2013). Reductions in speech intelligibility are a prominent feature of dysarthria (Darley, Aronson, & Brown, 1969). Studies suggest that even children who do not have clinical signs of dysarthria but have a CP diagnosis have reduced intelligibility relative to typically developing peers (Hustad, Sakash, Broman, & Rathouz, 2018; Hustad, Schueler, Schultz, & DuHadway, 2012).

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Intelligibility develops over time in all children. There have not been systematic large-scale studies establishing objective, empirical benchmarks for intelligibility development by age in typical children. However, available data across studies obtained with different methodologies suggest that typical children are likely to have intelligibility at about 90% for single words between 5 and 7 years of age (Wild, Vorperian, Kent, Bolt, & Austin, 2018). Similarly, 4- to 5-year-old children might be expected to have intelligibility for single words up to about 75%–80% on average (Hustad et al., 2012; Wild et al., 2018).

In a recent study, we examined longitudinal development of single-word intelligibility among a heterogeneous group of children with CP to provide an initial descriptive account of change over time (Hustad, Sakash, Natzke, Broman, & Rathouz, 2019). Findings revealed that, across all children with CP, half attained 25% intelligibility by 36 months; in addition, half attained 50% intelligibility by 48 months. Early intelligibility was highly predictive of later intelligibility. For example, the younger children were when they crossed the 25%, the 50%, or the 75% intelligibility threshold, the higher the maximum achieved intelligibility at 96 months. Children with CP as a group had reduced speech intelligibility at 96 months, with a median intelligibility of 73% and none of the children reaching 100% intelligibility. Growth in intelligibility development was most rapid from 3 to 5 years of age. It is noteworthy that children with CP were still growing in their intelligibility through 96 months, but that the rate of growth after 84 months of age was greatly reduced. Most of these findings are different than what might be expected for typically developing peers. However, given the range of abilities represented in the broad sample of children with CP, the disparity from typical development is not surprising.

Children with CP are heterogeneous in their specific disability profiles. Children can have a wide range of abilities in speech motor, language, and cognitive domains, which can yield different clinical presentations and functional ability profiles. Classification into subgroups (or profile groups)—smaller units with common features—is an important means to reduce heterogeneity, leading to a better understanding of manifestations of the disorder and enabling comparison among subgroups. Valid subgroups also allow for prediction of differential outcomes and for the systematic development and evaluation of interventions tailored to specific patterns of impairment. Efforts to develop and validate classification tools in the broader realm of CP research have been highly successful and include classification of underlying neuropathology, gross motor function, and fine motor function (Bax, Goldstein, Rosenbaum, & Levinton, 2005; Bax et al., 2006; Eliasson et al., 2006; Himmelman, Beckung, Hagberg, & Uvebrant, 2006; Pakula, Van Naarden Braun, & Yeargin-Allsopp, 2009; Rosenbaum, Palisano, Bartlett, Galuppi, & Russell, 2008; Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). In particular, data-inductive longitudinal study of gross motor development has led to important advances in our understanding of rates and limits of change and our ability to predict gross motor outcomes in children with CP (Hanna, Bartlett, Rivard, & Russell, 2008; Rosenbaum et al., 2002; E. Wood & Rosenbaum, 2000). Such information has important clinical, epidemiological, and theoretical implications for understanding and treatment of CP.

There are also several classification systems focused on speech and communication, including the Communication Function Classification System (Hidecker et al., 2011), the Viking Speech Scale (Virella et al., 2016), and the Functional Communication Classification System (Caynes et al., 2019). One classification model for considering speech and language ability profiles in children with CP was developed in our laboratory. Our paradigm, which we refer to as the *speech-language profile group* (SLPG) model, is based on foundations regarding the nature of CP and the co-occurrence of deficits (Bax et al., 2005, 2006; Odding, Roebroeck, & Stam, 2006; Rosenbaum et al., 2007). The SLPG model has two major dimensions: speech motor ability and language/cognitive ability. Because all individuals with CP have a movement disorder, but not all have speech motor involvement, the first level of differentiation is the presence or absence of clinical speech motor involvement. Children with no speech motor impairment (NSMI) constitute their own group. Within those who have speech motor involvement, children are further differentiated into those who are unable to produce speech and those who are able to speak but who have speech motor impairment (SMI). Finally, the model differentiates among children based on the presence or absence of language/cognitive deficits: those with SMI and typical language comprehension (SMI-LCT) and those with SMI and language comprehension impairment (SMI-LCI). Those who are unable to produce speech constitute their own group (anarthric). Earlier work provided empirical support for our SLPG model based on quantitative measures of speech and language (Hustad, Gorton, & Lee, 2010). In our more recent work, we have focused on validating the model (Hustad, Oakes, McFadd, & Allison, 2016) and on examining speech and language outcomes by profile group (Darling-White, Sakash, & Hustad, 2018; Hustad et al., 2017, 2018; Hustad, Sakash, Broman, & Rathouz, 2019; Lee, Hustad, & Weismer, 2014; Sakash, Broman, Rathouz, & Hustad, 2018).

In our previous study examining intelligibility growth in children with CP, we considered all children as one undifferentiated group. The goal of the present study was to refine our understanding of growth in speech intelligibility development among children with CP by examining the extent to which children with different communication profiles showed different intelligibility growth trajectories and different outcomes. To do this, we reanalyzed data from Hustad, Sakash, Natzke, et al. (2019) using statistical models that allowed us to quantify growth by SLPG. A key goal was to determine whether the inclusion of profile groups in our growth models improved prediction of speech intelligibility development outcomes relative to the aggregate findings from our earlier work. Results will allow us to consider the specific ways in which children with different profiles develop speech intelligibility, which in turn will help refine intervention decision making based on a child's specific clinical presentation. We addressed the following specific research questions:

1. How does the distribution of ages at which children reach 25%, 50%, and 75% intelligibility thresholds for single words differ across profile groups?
2. Are there differences in the average ages at which children have the greatest growth in single-word intelligibility based on profile group membership? Is there variability in age of greatest growth within profile groups? Are there differences in the rate of growth by profile group?
3. Are there differences across profile groups in maximum attained intelligibility (by or around 96 months of age)? How much heterogeneity is there in this end point across children within profile group?
4. Are there differences in how well single-word intelligibility at 36 months predicts single-word intelligibility at 96 months for the different profile groups? In what ways does profile group information improve the 96-month prediction?

We hypothesized that children in the NSMI group would show a distinct intelligibility advantage over children in the SMI groups because of the relative integrity of their speech motor abilities. Specifically, we expected that children in the NSMI group would reach intelligibility thresholds earlier, have the greatest intelligibility growth earlier, and reach a higher maximum intelligibility. Similarly, we expected that children with SMI-LCT would have an advantage over their peers with language comprehension impairment with regard to age of crossing thresholds, age of greatest growth, and maximum intelligibility owing to the added detrimental impact of comorbid language impairment and its potential association with memory and in some cases cognitive deficits, which might be expected to negatively interact with speech motor abilities. We anticipated that prediction of 96-month intelligibility outcomes would be improved with the addition of profile group membership and that 36-month intelligibility data would predict different intelligibility outcomes based on profile group membership.

Method

Participants

A cohort of 68 children with CP (32 girls, 36 boys), along with measurement materials and procedures, have been described in an earlier companion study (Hustad, Sakash, Natzke, et al., 2019). Briefly, children met the following criteria: (a) age between 18 and 60 months at initial enrollment; (b) medical diagnosis of CP; (c) hearing within normal limits as documented by either formal audiological evaluation or distortion product otoacoustic emission screening; (d) able to produce speech, which was operationally defined as the ability to repeat single words in an elicitation task; (e) contributed at least two longitudinal data points in which they produced speech; and (f) no codiagnosis of autism spectrum disorder. Each child contributed two to 13 data points, for a total of 564 data points, yielding a mean of 8.3 (*SD* = 2.6) and median of 9 data points per participant. All children were from homes where American English was the primary language and were born in the United States between 2000 and 2009. Demographic information, including CP diagnosis, is presented in Table 1. Table 2 provides a summary of how many children were enrolled in speech/language therapy during each visit represented in this article. Potential implications of therapy are addressed in the context of findings from this study and are elaborated in the Discussion section.

Two different nondisabled adult listeners were quasi-randomly assigned to orthographically transcribe the speech of each child and each visit (564 visits across the 68 different children × 2 listeners per visit = 1,128 listeners). Each listener heard only one child at one visit producing all stimulus material. Inclusion criteria for listeners were as follows: (a) pass pure-tone hearing screening administered via headphones at 25 dB HL for 250 Hz, 500 Hz, 1 kHz, 4 kHz, and 6 kHz bilaterally; (b) between 18 and 45 years of age; (c) no more than incidental experience listening to or communicating with persons having communication disorders; (d) native speaker of American English; and (e) no identified language, learning, or cognitive disabilities per self-report. Listeners were 305 males and 825 females. The mean age of listeners was 20.8 (*SD* = 5.6) years.

Materials and Procedures

Children participated in a standard speech and language assessment protocol administered by a research speech-language pathologist (SLP) in a sound-attenuating suite for each session. Methodological details regarding speech stimuli, elicitation procedures, data collection from listeners, and intelligibility measures are provided in Hustad, Sakash, Natzke, et al. (2019) and are thus abbreviated here.

Acquisition of Speech Samples From Children

Children produced speech stimuli from the Test of Children's Speech Plus (TOCS+; Hodge & Daniels, 2007) at each visit. Speech samples from children were recorded using a digital audio recorder (Marantz PMD570) at a 44.1-kHz sampling rate (16-bit quantization), with a condenser studio microphone (Audio-Technica AT4040) positioned next to each child, approximately 18 in. from the child's mouth.

Acquisition of Intelligibility Data

Digital recordings of children's speech were edited to remove any extraneous noises and peak amplitude normalized for playback to listeners. In a sound-attenuating suite, listeners utilized in-house software to complete an experimental task involving orthographic transcription of children's speech. Speech samples were presented in a sound field with peak audio output levels calibrated to approximately 75 dB SPL from where listeners were seated. Individual words produced

Table 1. Demographic and clinical characteristics of children with cerebral palsy (CP).

Characteristic	NSMI (*n* = 23)	SMI-LCT (*n* = 31)	SMI-LCI (*n* = 14)
Number of visits, *M* (*SD*)	8.7 (2.5)	8.8 (2.2)	6.5 (2.6)
Number of visits, *Mdn*	9.0	9.0	6.0
Male:female ratio	18:5	11:20	7:7
CP type			
Spastic			
Diplegia	6	6	1
Hemiplegia (left)	9	4	1
Hemiplegia (right)	4	8	4
Triplegia	0	1	1
Quadriplegia	0	5	1
Unknown	1	1	0
Dyskinetic	0	1	0
Ataxic	1	3	1
Mixed	0	0	1
Hypotonic	0	1	0
Unknown	2	1	4
GMFCS level			
I	20	13	6
II	3	4	1
III	0	6	1
IV	0	7	4
V	0	1	2

Note. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment; GMFCS = Gross Motor Function Classification System (Palisano et al., 1997).

by children were randomized for each listener and presented in isolation. Listeners heard each word produced by the child one time. They were instructed that children would be producing real words and to take their best guess if they were unsure as to what the child said. Data collection from listeners took approximately 30 min per listener.

Analysis of Speech Intelligibility Data

For each child and each visit, two independent listeners generated typed orthographic transcriptions of each stimulus word. We used two listeners per child because our previous work has shown that it is necessary to use more than one listener per child to ensure that intelligibility scores reflect listener variability known to be present in intelligibility tasks of this nature (Hustad, Oakes, & Allison, 2015). We used independent listeners for each child and visit to minimize the potential for learning effects associated with hearing multiple children and/or the same stimuli produced by different speakers. We wanted listener data to reflect naïve listeners without experience; therefore, listeners were only exposed to the speech of one child at one visit. This study was large in scope, comprising 564 sets of speech samples across children and visits. We used only two listeners per child (1,128 listeners) because of cost and time required for collecting intelligibility data from additional listeners per child.

Table 2. Number of children by profile group reporting a given amount of therapy across visits (*N* = 68 children).

Percentage of visits reporting therapy	NSMI	SMI-LCT	SMI-LCI
0–25	16	5	0
26–50	4	4	0
51–75	2	10	3
76–100	1	12	11

Note. Therapy information was not provided for 10 visits across a total of six children. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment.

Intelligibility scores by child and visit were obtained by counting the number of words that were an exact phonemic match to the target word produced by the child for each listener. The total number of words transcribed correctly by each of the two listeners per child was averaged and then divided by 38 (the number of words produced by each child) to yield a mean intelligibility score expressed as a proportion for each child and each visit.

We calculated interrater reliability on intelligibility scores with the intraclass correlation coefficient estimated using the irr R package (Version 0.84.1; Gamer, Lemon, Fellows, & Singh, 2019). We used a single-score, absolute-agreement, two-way random effects model, and we found strong agreement for the 564 listener pairs, intraclass correlation coefficient = .955, 95% CI [.948, .962]. Moreover, the average difference between the two listeners of each child for each visit was 6.19 percentage points (5.1 *SD*s). These values are within an acceptable range (Lee et al., 2014).

Receptive Language Assessment

The following measures were administered, depending on the child's age, developmental level, and motor skill profile: the Test for Auditory Comprehension of Language–Third Edition (TACL-3; Carrow-Woolfolk, 1999), the Preschool Language Scale–Fourth Edition (PLS-4; Zimmerman, Steiner, & Pond, 2002), and the Peabody Picture Vocabulary Test–Fourth Edition (Dunn & Dunn, 2007).

Rubric for selection of language comprehension measure for each longitudinal session was as follows. Children under the chronological age of 3 years received the PLS-4, which assesses earlier acquired skills with portions administered via parent interview. We attempted to administer the TACL-3 for all children over the chronological age of 3 years. The TACL-3 requires limited motor skills, as response options are presented in a field of three discrete pictures, which can be selected using manual direct selection or partner-directed scanning. However, if a child was not able to participate in administration of the TACL-3, the PLS-4 was administered. The Peabody Picture Vocabulary Test–Fourth Edition was administered to children who could not tolerate the longer TACL-3 but who could participate in picture-pointing tasks via manual direct selection or partner-directed scanning. Standard administration procedures were adapted to enable participation in testing for items involving manual manipulation. Instructions in the technical manuals were followed for setting up adaptations.

Standard scores on each test were obtained following respective technical manuals. Note that, even in instances where standardization procedures for test administration were violated, we proceeded with use of standard scores. Standard scores were used to make binary decisions regarding the presence or absence of language comprehension impairment. Language comprehension impairment was defined based on standard scores that were at least 1.5 *SD*s below age expectations per the technical manual of respective tests (Carrow-Woolfolk, 1999; Dunn & Dunn, 2007; Zimmerman et al., 2002).

Classification Into Profile Groups

Children were classified into SLPGs following our earlier work (Hustad et al., 2010, 2016) on the basis of data obtained from their visit closest to 48–54 months of age. We used a retrospective classification approach where we looked backward in time at early behavior, given knowledge of later outcomes because our studies suggest that children with CP cannot be readily classified using the SLPG paradigm prior to 4 years of age (Hustad, Allison, McFadd, & Riehle, 2014). In particular, diagnosis of SMI is challenging prior to the age of 4 years because children with CP tend to be delayed in the onset of speech and in their early speech development and because features of early typical speech development overlap with features of SMI (i.e., reduced rate of speech, reduced intelligibility, omissions, substitutions, and distortions of speech sounds) in young children. However, at the age of 4 years, we have been able to reliably diagnose the presence or absence of SMI in children with CP (Hustad et al., 2010, 2016). Because of the stable nature of the neurological involvement that underlies CP, children do not tend to outgrow SMI, although speech characteristics do change with development. We have used this retrospective classification approach in previous work (Hustad et al., 2018), and findings have provided useful information regarding how children get to their later classification outcomes. Note that in this article, we use the same children and data set as the earlier Hustad, Sakash, Natzke, et al. (2019) study, with the exception of one child who had two data collection sessions prior to the age of 4 years and therefore could not be classified for this study. This child was omitted from the analyses.

Children were independently classified by two SLPs based on judgment of the presence or absence of SMI and the presence or absence of language impairment. Children who were classified as having speech motor involvement (SMI), by definition, had clinical evidence of dysarthria. SMI was determined through clinical observation of the presence or absence of common features of dysarthria, including facial asymmetry; drooling; hypernasality; short breath groups; breathy, harsh, or wet vocal quality; imprecise articulation; and consonant or vowel substitutions, distortions, or omissions that were not age appropriate. Perceptual judgments were made from spontaneous speech samples between the child and a parent or the child and a clinician as well as speech samples obtained from production of the TOCS+ (Hodge & Daniels, 2007) stimuli in a delayed imitation task. Note that intelligibility data were not used in the classification of children. All children in the SMI groups had evidence of dysarthria as their primary speech motor disorder.

To characterize language impairment, we considered language comprehension abilities only. Current and previous research suggests that language comprehension abilities are a reasonable proxy for cognitive abilities as indicated by studies showing similar scores on both measures (Cole, Mills, & Kelley, 1994; Kilbride, Thorstad, & Daily, 2004) and recent work showing strong convergence between cognitive and receptive language deficits in children with CP (Soriano & Hustad, 2019). We did not use expressive language measures because the majority of the children in this study had SMI, which effectively served as a barrier to accurate characterization of expressive language features. Language comprehension impairment was operationally defined as standard scores that were at least 1.5 *SD*s below age expectations per the technical manual of respective tests (Carrow-Woolfolk, 1999; Dunn & Dunn, 2007; Zimmerman et al., 2002).

Children who were classified as having NSMI (n = 23 children) had no clinical evidence of speech or language impairment based on clinician observation. Note that, in making the classification of NSMI, we only considered clinical impressions of speech production abilities made by experienced research SLPs. We did not examine test scores or other speech and language assessment measures; thus, children in this group may have had subtle or borderline speech and language difficulties that were separate from speech motor abilities, including developmental articulation disorders and/or expressive language challenges. Detailed analyses of speech and language abilities in the NSMI group were beyond the scope of this article but are an important direction for future research.

Children who had SMI and language comprehension that was within age expectations were classified as SMI-LCT (*n* = 31). Children who had SMI and language comprehension impairment were classified as SMI-LCI (*n* = 14 children). Children who were classified as anarthric had severe SMI, producing fewer than five words or word approximations based on parent report and based on clinician observation during the data collection session. These children were omitted from this study as they were not able to produce TOCS+ stimuli for speech intelligibility measurement.

Reliability of our clinical classification of children has been documented in previous studies (Hustad et al., 2010, 2016). Classification agreement between the two SLP raters was 100%.

Statistical Analyses

Recently, we developed and justified an analysis approach to longitudinal growth trajectory measures of single-word intelligibility via nonlinear random effects models (Davidian, 2017; Davidian & Giltinan, 1995; Hustad, Sakash, Natzke, et al., 2019). The approach dealt with several challenging issues with this cohort and yielded interpretable results and novel insights. The approach was based on a balance of descriptive plots of the data with models via a modified logistic function of the form in Equation 1, with parameters asymptote, midpoint (inflection point), and scale (rate of change).

$$f(t) = \frac{\text{asymptote}}{1 + e^{\left(\frac{\text{midpoint}-t}{\text{scale}}\right)}} \tag{1}$$

In this model, the mean response (average intelligibility) for each child is given by the function $f(t)$, where t represents the child's age in months. Additionally, the functional form is characterized by three key parameters: the asymptote or the maximum/plateau value on the curve; the midpoint, which is the value of time age t when the intelligibility growth is the most rapid; and a numeric scale parameter representing the overall growth factor for the curve. Of these features, the asymptote and midpoint are directly interpretable. The asymptote represents the maximum value of the growth curve. The asymptote estimates were within 1 percentage point of the estimated intelligibility at 96 months. The midpoint parameter of the intelligibility growth curve defines an inflection point in the curve's growth. Prior to the midpoint, growth accelerates away from the baseline of 0% intelligibility, and after the midpoint, growth slows down and intelligibility plateaus as it approaches the curve's maximum intelligibility asymptote. The midpoint marks the point when the rate of growth is the greatest. In general, the scale parameter is not directly interpretable because the growth rate on the curve depends on the asymptote and distance from the midpoint. We can use the scale and asymptote, however, to compute the maximal rate of growth: growth at the midpoint is equal to asymptote / (4 × scale) intelligibility percentage points per month. Importantly, each child was accorded his or her own value for the asymptote and for midpoint via the random effects structure of the model. We used the fitted model and the estimated values of the asymptote and midpoint to estimate the plateau intelligibility that each child will reach and also each child's age at maximum growth. Analyses were performed using the R programming language (Version 3.5.2; R Core Team, 2018). Nonlinear mixed effects models were fitted using the lme4 R package (Version 1.1.21; Bates, Mächler, Bolker, & Walker, 2015).

In the new analysis presented here, average trajectories were allowed to vary across the three SLPGs by estimating different values of the asymptote, midpoint, and scale for each group. In addition, by testing for any differences in these three values across the three groups, we are able to test, globally, for any differences in mean trajectories across the three groups. As follow-on analyses to this global hypothesis test, we can conduct tests between any of the three pairs of groups and/or in values of any of the three parameters among the three groups. Assuming such differences are detected, we can then quantify differences in the median ages at which 25%, 50%, and 75% intelligibility thresholds are crossed as a function of group membership. These tests and estimates provide quantitative responses to Research Questions 1 through 3.

The fourth question can be addressed by using the fitted model, in which the groups vary by asymptote, by midpoint, and by scale to simulate children from each group and then to predict mean 96-month value as a function of 36-month value observed in a clinical setting. This approach is similar to our earlier analysis (Hustad, Sakash, Natzke, et al., 2019). We then examine and quantify the degree to which the additional information of SLPG membership further enhances the 96-month prediction. This addresses the critical question as to whether broad clinical assessment of both speech and language comprehension is useful as an indicator of future speech impairment.

Our simulation procedure was as follows. We simulated new children by randomly resampling from the observed children from our data set. Each observed child had two conditional posterior modes from the model's random effects; these were the by-child estimates of the asymptote and midpoints. Each child also had a conditional posterior variance–covariance matrix for the asymptote and midpoint estimates. We sampled from the multivariate normal distributions defined by these conditional modes and variance–covariance matrices to simulate new child-specific asymptotes and midpoints. We then added the group average growth curve features (i.e., the fixed effects) and computed the intelligibility averages at 36 and 96 months. Finally, we jittered these averages by sampling from the model's residual error. This procedure incorporated uncertainty in group heterogeneity (by resampling children), incorporated uncertainty in child-level estimates of growth curve features (by simulating asymptotes and midpoints for resampled children), and incorporated uncertainty from overall residual error. We simulated 1,000,000 children using this procedure.

Results

Question 1: How does the distribution of ages at which children reach 25%, 50%, and 75% intelligibility thresholds for single words differ across children's profile groups?

Figures 1–3 show the observed intelligibility growth trajectories and model-estimated intelligibility thresholds for each of the profile groups. The dot plots and box plots below the trajectories visualize the ages when each child in each group crosses the 25%, 50%, and 75% thresholds.

All of the children in the NSMI group (see Figure 1) reached the 25% and 50% intelligibility thresholds. For the 25% intelligibility threshold, model results indicate that 25% of children reached the threshold by 25 months of age, 50% of the children reached the threshold by 29 months of age, 75% of children reached the threshold by 32 months of age, and 100% of children reached the threshold by 41 months of age. For the 50% intelligibility threshold, model results revealed children crossed this threshold about 12 months later. In particular, 25% of children reached the 50% threshold by 36 months, 50% of children crossed the threshold by 40 months, 75% of children reached the 50% threshold by 43 months, and 100% of children reached the threshold by 54 months. Finally, children in the NSMI group crossed the 75% intelligibility threshold about 15 months later. Specifically, 25% of children reached 75% intelligibility by 50 months of age, 50% reached it by 55 months of age, and 75% reached it by 60 months of age. By 100 months of age, 87% of children in the group had reached 75% intelligibility.

Figure 1. Observed trajectories plus predicted ages crossing for 25%, 50%, and 75% single-word intelligibility thresholds for children with cerebral palsy and no speech motor impairment (NSMI). Box plots below the trajectory plot show the distribution of the predicted ages at which each child's trajectory crossed the 25%, 50%, and 75% intelligibility thresholds. Predicted ages that fall outside the 20- to 100-month range are plotted at the extremes of these ranges. Children's colors are ordered by their predicted maximum intelligibility (i.e., the by-child random asymptote).

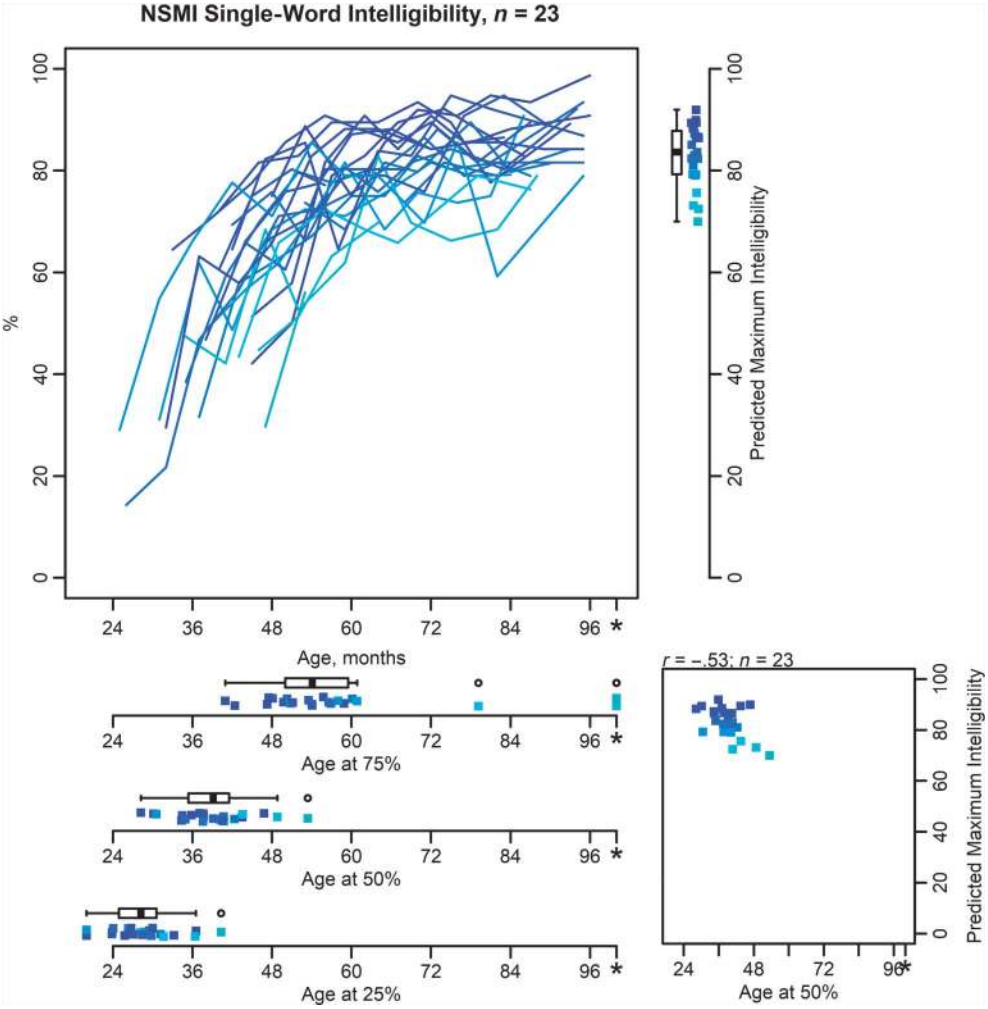
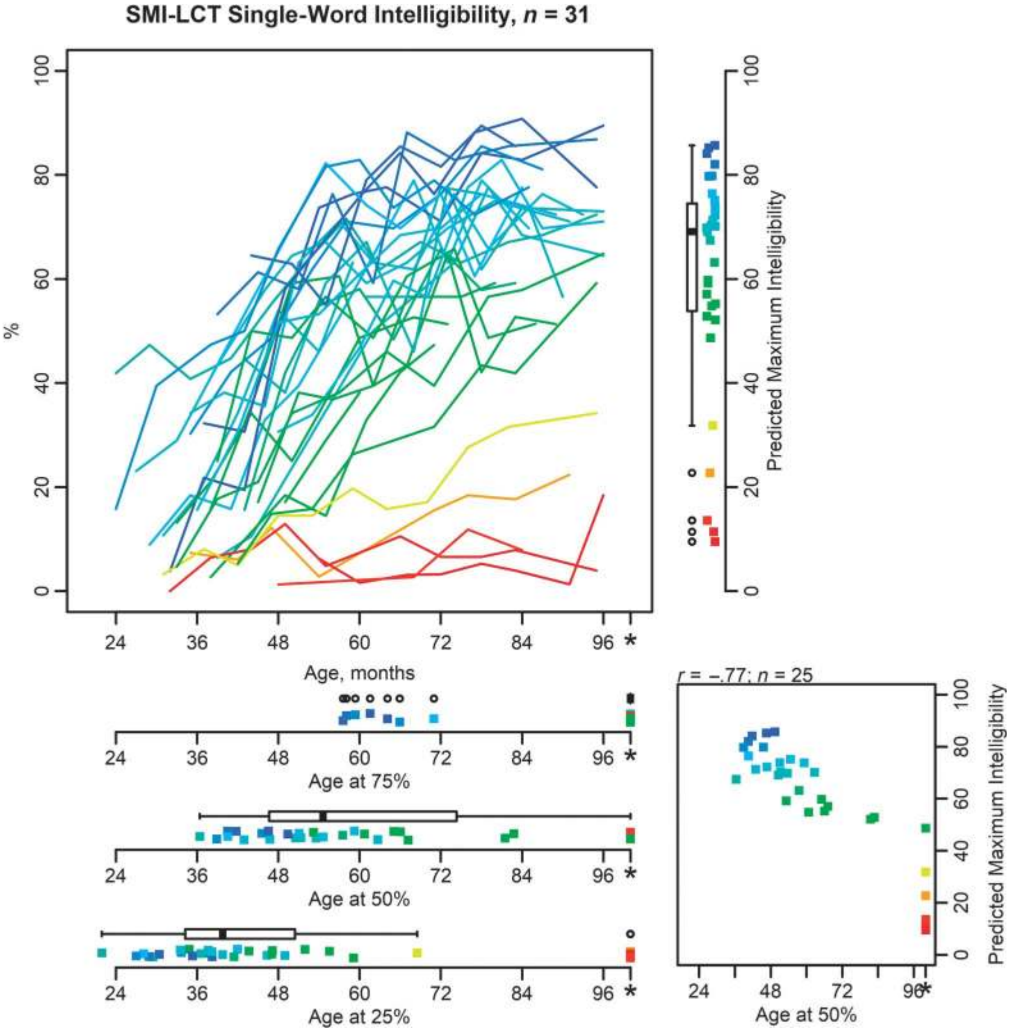


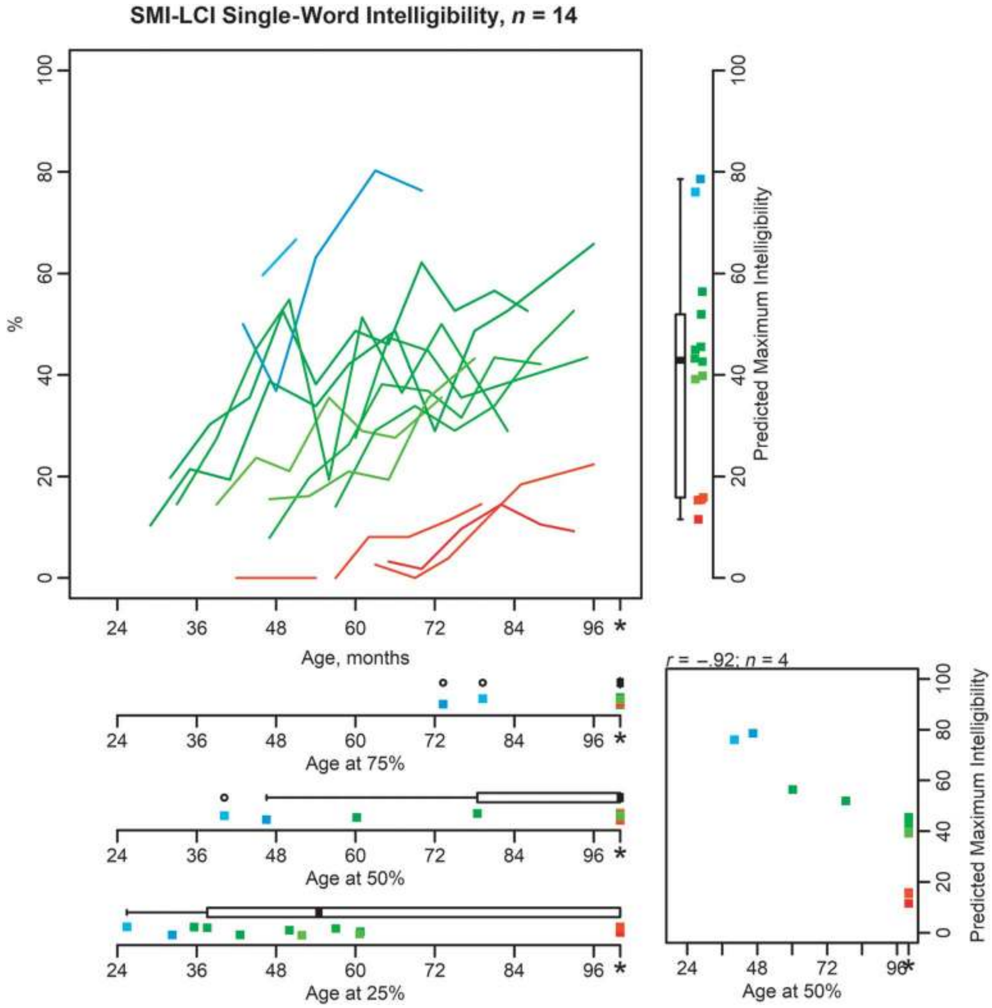
Figure 2. Observed trajectories plus predicted ages crossing for 25%, 50%, and 75% single-word intelligibility thresholds for children with cerebral palsy, speech motor impairment, and language comprehension that is typically developing (SMI-LCT). Box plots below the trajectory plot show the distribution of the predicted ages at which each child's trajectory crossed the 25%, 50%, and 75% intelligibility thresholds. Predicted ages that fall outside the 20- to 100-month range are plotted at the extremes of these ranges. Children's colors are ordered by their predicted maximum intelligibility (i.e., the by-child random asymptote).



For the SMI-LCT group (see Figure 2), there were not any intelligibility thresholds that all of the children in the group crossed. Based on the modeled intelligibility growth trajectories, 13% of the children in this group did not cross the 25% intelligibility threshold by 100 months of age; this subset of children can be seen in Figure 2 with observed intelligibility scores that were relatively flat over time. For the 25% intelligibility threshold, model results indicated that 25% of children crossed the threshold by 34 months of age, 50% of children crossed the threshold by 40 months of age, and 75% of children crossed the threshold by 52 months of age. For the 50% intelligibility threshold, children in the SMI-LCT group crossed the threshold about 12–30 months later than the 25% threshold. Specifically, 25% of children crossed the 50% intelligibility threshold by 47 months, 50% of children crossed this threshold by 55 months, and 75% of children crossed this threshold by 82 months. Although 81% of children in this group were expected to reach 50% intelligibility by 100 months, just 23% were expected to reach 75% intelligibility by 100 months.

For the SMI-LCI group (see Figure 3), again, there were not any intelligibility thresholds that all of the children crossed. In fact, the estimated growth trajectories for 29% of the children did not cross the 25% intelligibility threshold by 100 months of age. This subset of children can be seen in Figure 3 as a set of relatively flat lines. For the 25% intelligibility threshold, model results indicated that 25% of children crossed the threshold by 38 months whereas 50% of children crossed it by 52 months. For the 50% intelligibility threshold, 25% of children crossed the threshold at 79 months. Just 29% and 14%

Figure 3. Observed trajectories plus predicted ages crossing for 25%, 50%, and 75% single-word intelligibility thresholds for children with cerebral palsy, speech motor impairment, and language comprehension that is impaired (SMI-LCI). Box plots below the trajectory plot show the distribution of the predicted ages at which each child's trajectory crossed the 25%, 50%, and 75% intelligibility thresholds. Predicted ages that fall outside the 20- to 100-month range are plotted at the extremes of these ranges. Children's colors are ordered by their predicted maximum intelligibility (i.e., the by-child random asymptote).



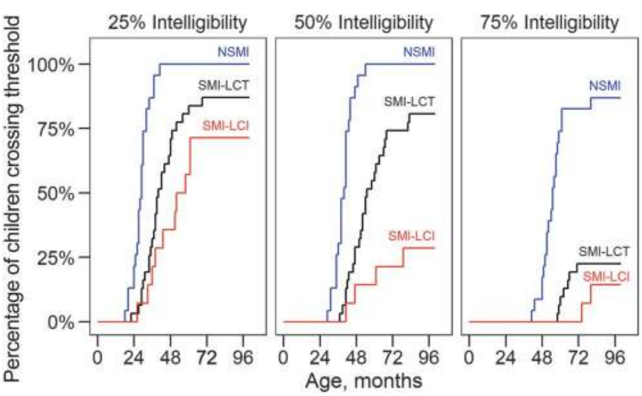
of children in this group were expected to reach 50% and 75% intelligibility by 100 months, respectively. Figure 4 shows comparative results by profile group, illustrating the cumulative incidence of intelligibility threshold by age.

Question 2: Are there differences in the average ages at which children have the greatest growth in single-word intelligibility based on profile group membership? Is there variability in age of greatest growth within profile groups? Are there differences in the rate of growth by profile group?

Histograms in Figure 5 show the distribution of the ages of maximum intelligibility growth in each group. The estimated ages of maximum growth varied considerably within each group. In the NSMI group, the ages were tightly clustered around 3 years of age. The ages ranged from 26 to 45 months, with an interquartile range of 32–38 months. The ages of maximum growth for the SMI-LCT group covered a wider range than the NSMI group, 27–66 months, and 17 of the 31 children had estimated midpoints after 45 months (the latest age estimated in the NSMI group). The SMI-LCI group covered a similar range as the SMI-LCT group, 33–65 months. In this case, most children (nine of 14) had ages of maximum growth outside the range observed in the NSMI group.

Table 3 reports the estimated group means for the age of maximum growth (midpoint), and Table 4 reports the estimated differences of the group means for the age of maximum growth (midpoint). Broadly speaking, the average intelligibility in the NSMI group grew most quickly around 3 years of age, while the two SMI groups showed the greatest growth closer to 4 years of age. Specifically, based on model results, growth was steepest for the NSMI group at 35.2 months, 95% CI [29.4, 41.0]. At this age, children in

Figure 4. Cumulative incidences of intelligibility thresholds by age. The lines in each panel show the cumulative percentage of children in each group who reached the intelligibility threshold at each age. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment.



the group had an estimated average growth rate of 2.36 intelligibility percentage points per month, 95% CI [1.91, 2.80]. For the SMI-LCT group, the estimated average age for maximum growth was 46.4 months, 95% CI [41.4, 51.4], with an estimated average growth rate of 1.66 intelligibility percentage points per month, 95% CI [1.41, 1.91]. For the SMI-LCI group, the estimated average age of maximum growth was 49.1 months, 95% CI [40.1, 58.2], with an estimated average growth rate of 1.00 intelligibility percentage points per month, 95% CI [0.61, 1.36]. Children in the NSMI group showed their steepest growth significantly earlier than the SMI-LCT group, $\mathrm{diff}_{\mathrm{NSMI-LCT}} = -11.2$, $SE = 2.8$, $z = -4.0$, $p < .001$, and the SMI-LCI group, $\mathrm{diff}_{\mathrm{NSMI-LCI}} = -13.9$, $SE = 3.9$, $z = -3.57$, $p < .001$. There was not a significant difference between the points of steepest growth for the SMI-LCT and SMI-LCI groups, $\mathrm{diff}_{\mathrm{LCT-LCI}} = -2.7$, $SE = 3.7$, $z = -0.73$, $p = .46$.

Question 3: Are there differences across profile groups in maximum attained intelligibility (by or around 96 months of age)? How much heterogeneity is there in this end point across children within profile group?

Box plots in the right margins of Figures 1–3 show the predicted maximum intelligibility value for children in each group. Overall, there was considerable variability in the predicted maximum intelligibility values by group. For the NSMI group, the estimated maximum intelligibilities were tightly clustered, ranging from 70% to 92%. The two groups of children with SMI showed much more variability. In the SMI-LCT group, estimated maximum intelligibilities ranged from 10% to 86%. This range was influenced by five outlying children whose estimated maximum intelligibilities were less than 40%. Most of the children (75%) in the group had estimated intelligibilities of 54% or greater. A similar pattern occurred in the SMI-LCI group, whose estimated maximum intelligibilities ranged from 12% to 79%. There were four outlying children in the SMI-LCI group who had estimated intelligibilities of less than 20%, but most of the children (71%) had estimated intelligibilities of 39% or greater. Table 3 reports the estimated group means for the maximum attained intelligibility (asymptote), and Table 4 reports the estimated differences in the group means.

For the NSMI group, the estimated average maximum intelligibility was 82.0%, 95% CI [71.3, 92.6]. The NSMI group had a significantly greater estimated maximum intelligibility value than the SMI-LCT group, $\mathrm{diff}_{\mathrm{NSMI-LCT}} = 21.1$, $SE = 5.1$, $z = 4.13$, $p < .001$, and the SMI-LCI group, $\mathrm{diff}_{\mathrm{NSMI-LCI}} = 39.5$, $SE = 6.6$, $z = 5.99$, $p < .001$. The SMI-LCT group had an estimated average maximum intelligibility of 60.8%, 95% CI [51.6, 70.1], and the SMI-LCI group had an estimated average maximum intelligibility of 42.4%, 95% CI [27.7, 57.2]. The estimated maximum intelligibility for the SMI-LCT group was significantly greater than that for the SMI-LCI group, $\mathrm{diff}_{\mathrm{LCT-LCI}} = 18.4$, $SE = 6.3$, $z = 2.91$, $p = .004$.

Question 4: Are there differences in how well single-word intelligibility at 36 months predicts single-word intelligibility at 96 months for the different profile groups? In what ways does profile group improve the 96-month prediction?

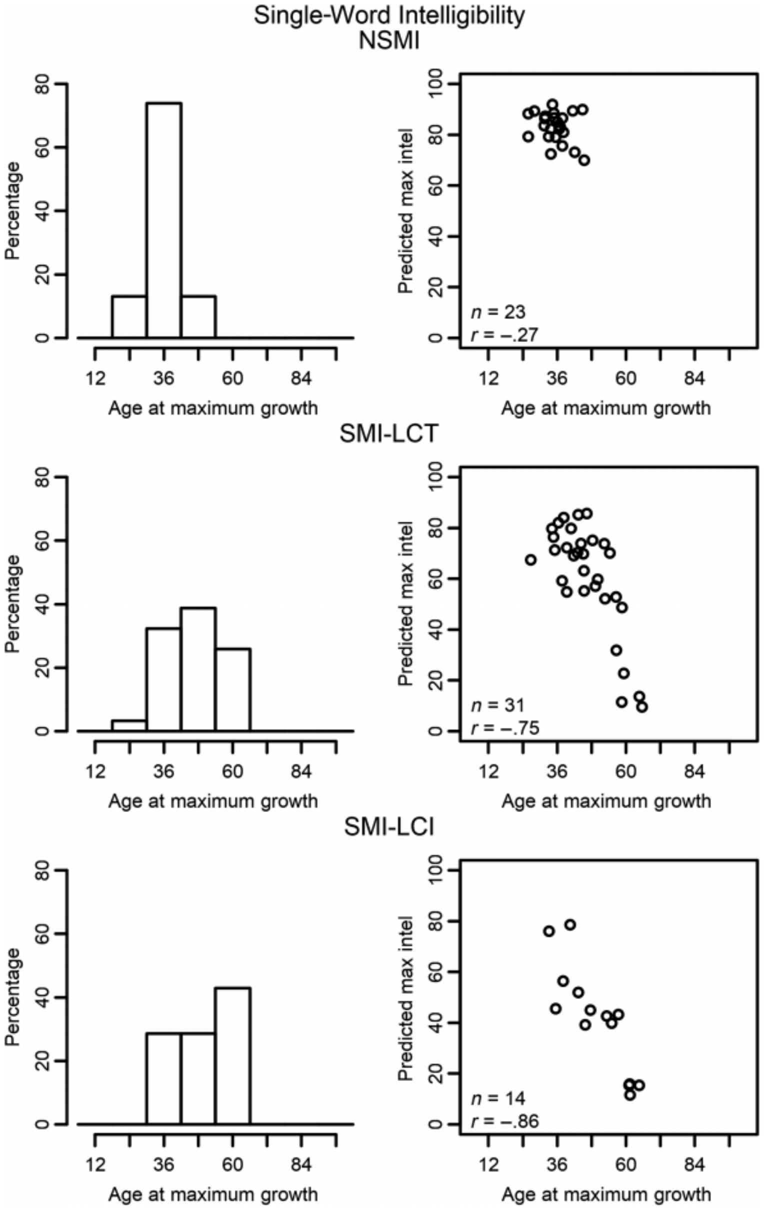
We examined how intelligibility at 36 months predicted intelligibility at 96 months for simulated participants described previously. We fit a generalized additive model with cubic regression splines using the mgcv R package (Version 1.8.26; S. N. Wood, 2017) to estimate the nonlinear relationship between 36-month and 96-month intelligibility. The model also included group as a predictor. The baseline model with only 36-month intelligibility had an adjusted $R^2 = .78$. Adding group as a predictor increased the amount of explained variance slightly, adjusted $R^2 = .84$ (see Figure 6). Based on model comparisons, group membership provides a small predictive benefit over 36-month intelligibility alone.

Figure 6 shows the predictions from the second model. To prevent overplotting, just 300 of the simulated children are plotted from each group. The lines are the predicted average intelligibility at 96 months using the additive model with 36-month intelligibility and profile group. One feature to notice is the funnel on the left side of the figure. Given very low 36-month intelligibilities (horizontal axis), outcomes at 96 months (vertical axis) are highly uncertain. For 36-month intelligibilities in the 0%–10% range, the group average predictions ranged from 6% to 68%. The group differences in intelligibility become apparent for 36-month intelligibility scores that are higher than 10%. Children in the NSMI group are expected to have a 96-month intelligibility about 10 percentage points higher than children from the other two groups. The predictive value of 36-month intelligibility plateaus after about 35%. Differences in 36-month intelligibility for values over 35% do not predict substantially different outcomes in 96-month intelligibility (see Table 5 for the global tests of group differences).

Discussion

The purpose of this study was to refine our previously published (Hustad, Sakash, Natzke, et al., 2019) account of speech intelligibility development in children with CP on

Figure 5. Histograms and scatter plots of the distribution of age of maximum growth in speech intelligibility by profile group. Note that the histograms show the percentages of children in each group with maximum growth ages in each 12-month bin. The scatter plots show the correlation between age at maximum growth and model-predicted maximum intelligibility. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment.



the basis of a speech and language classification paradigm, which would allow us to separate children into profile groups based on the constellation of speech and language deficits observed at 4 years of age. In particular, we were interested in determining whether children with different speech and language impairment profiles differed in their speech intelligibility growth and whether profile group membership would improve our ability to predict longer term intelligibility outcomes at 8 years on the basis of data obtained at 36 months. This study revealed clear evidence that separating children into profile groups yields important differences regarding the age of crossing intelligibility thresholds, growth, attainment of intelligibility outcomes among subsets of children with CP, and prediction of intelligibility outcomes at 8 years on the basis of data from 3 years of age. Specifically, key findings from this study were as follows: (a) Children with CP who have NSMI have substantially different growth and better intelligibility outcomes than their peers who have SMI, (b) children who have SMI-LCT tend to have better outcomes but similar

Table 3. Estimated group means.

Curve feature	Group	Estimate	*SE*	95% CI
Asymptote (%)	NSMI	82.0	3.9	[71.3, 92.6]
	SMI-LCT	60.8	3.4	[51.6, 70.1]
	SMI-LCI	42.4	5.4	[27.7, 57.2]
Midpoint (months)	NSMI	35.2	2.1	[29.4, 41.0]
	SMI-LCT	46.4	1.8	[41.4, 51.4]
	SMI-LCI	49.1	3.3	[40.1, 58.2]
Scale	NSMI	8.7	0.8	[6.5, 10.9]
	SMI-LCT	9.1	0.6	[7.5, 10.8]
	SMI-LCI	10.8	2.1	[5.0, 16.6]
Rate (% per month at midpoint)	NSMI	2.4	0.2	[1.9, 2.8]
	SMI-LCT	1.7	0.1	[1.4, 1.9]
	SMI-LCI	1.0	0.2	[0.6, 1.4]

Note. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment.

growth as children with SMI-LCI, and (c) there seems to be a subset of children that cut across SMI-LCI and SMI-LCT groups who have severe speech motor involvement and show limited growth in intelligibility. These findings and their clinical implications are discussed in detail.

Different Growth and Different Outcomes for Children With NSMI Than Children With SMI

Children with NSMI crossed more intelligibility thresholds (i.e., 25% intelligibility, 50% intelligibility, 75% intelligibility) than their peers in the two SMI groups, and they did so at earlier ages (approximately a year prior to children in SMI groups). Notably, most children with NSMI crossed the 75% intelligibility threshold by about 5 years of age, a finding that is generally consistent with the very limited information on intelligibility development in typically developing children at this same age (Hodge & Gotzke, 2014). However, less than a quarter of children with SMI ever crossed this threshold. Findings from this study indicated that children with NSMI had mean intelligibility scores of 82% at 8 years of age, and none of the children reached 100% intelligibility in this study. Another recent study found that, at 5 years of age, speech intelligibility, speech rate, and intelligible words per minute in children with CP and NSMI were more like typical children than like children with SMI, but they were, in fact, differentiable from typical peers on all three measures (Hustad, Sakash, Broman, et al., 2019).

Table 4. Estimated differences of group means.

Curve feature	Comparison	Estimate	*SE*	*z*	*p*
Asymptote (%)	NSMI–LCT	21.1	5.1	4.13	< .001
	NSMI–LCI	39.5	6.6	5.99	< .001
	LCT–LCI	18.4	6.3	2.91	.004
Midpoint (months)	NSMI–LCT	–11.2	2.8	–4.00	< .001
	NSMI–LCI	–13.9	3.9	–3.57	< .001
	LCT–LCI	–2.7	3.7	–0.73	.464
Scale	NSMI–LCT	–0.4	1.0	–0.45	.654
	NSMI–LCI	–2.1	2.2	–0.92	.356
	LCT–LCI	–1.6	2.2	–0.75	.455

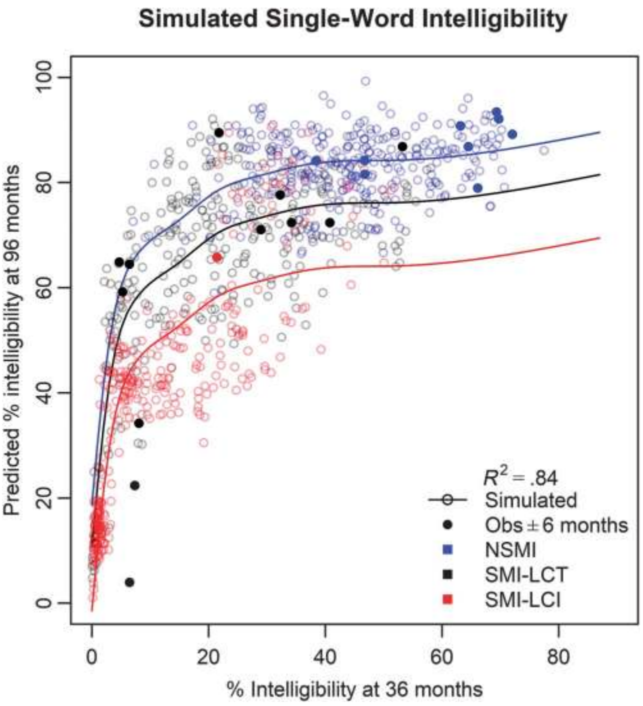
Note. NSMI = no speech motor impairment; LCT = language comprehension typical; LCI = language comprehension impairment.

The period of maximum growth for children in the NSMI group was earlier than for those in the SMI groups, shifting the age range down by 12 months and narrowing it (range: 24–54 months, $M = 36$). This finding refines our earlier work indicating that when all children with CP were pooled into one analysis, maximum growth occurred at 36–60 months of age (Hustad, Sakash, Natzke, et al., 2019). One potential reason for the earlier growth spurt among children with NSMI is the presence of fewer comorbidities and medical complexities than observed in the children with SMI. Specifically, the presence of more comorbidities and medical complexities in children with SMI may have served to increase the likelihood of delays in development, particularly in the very early years. In addition, children in the NSMI group grew at a faster rate, overall, than those in the SMI groups. This finding is consistent with work on receptive language development in which children with NSMI were found to have accelerated growth relative to both their peers with SMI and typically developing expectations (Hustad et al., 2018).

Intelligibility scores at 36 months for children in the NSMI group predicted outcomes that were about 10–20 percentage points better at 8 years of age than those predicted for the SMI groups. Prediction accuracy overall was improved when profile group information was included in our modeling, compared to earlier work in which profile groups were not incorporated. However, there is a question regarding whether the improvement in R^2 measures from .78 (Hustad, Sakash, Natzke, et al., 2019) to .84 (this study) is clinically meaningful.

Results showing better intelligibility outcomes for children with NSMI than for children in SMI groups are not surprising given that children with NSMI, by definition, do not have evidence of clinical speech motor involvement. However, the extent to which children with NSMI differ

Figure 6. Scatter plot showing prediction of 96-month intelligibility outcomes from 36-month single-word intelligibility data by profile group. Data were obtained using single-word intelligibility data simulations based on models depicted in Figures 1–3. Solid points represent data from observed children, and open points represent data from simulations. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment–language comprehension typical; SMI-LCI = speech motor impairment–language comprehension impairment.



from or are similar to typically developing peers on these same metrics is unknown. Parallel data are not available for typically developing children, but previous work suggests that children with NSMI may lag behind their typically developing peers in speech intelligibility at 4 years of age (Hustad et al., 2012). Further studies are needed to investigate the ways that children with NSMI are similar to and different from typical peers across the age span. If children with NSMI show delays in growth and intelligibility outcomes relative to typical peers, they may be candidates for speech therapy to normalize speech production abilities.

Table 5. Global tests of group differences.

Comparison	*F*	*df*	*p*
No group contrasts	8.31	6, 551	< .001
NSMI vs. LCT	7.73	3, 551	< .001
NSMI vs. LCI	14.69	3, 551	< .001
LCT vs. LCI	3.82	3, 551	.010

Note. NSMI = no speech motor impairment; LCT = language comprehension typical; LCI = language comprehension impairment.

Better Outcomes for SMI-LCT Than SMI-LCI but Similar Growth

Children with SMI had delayed acquisition of intelligibility thresholds (i.e., 25% intelligibility, 50% intelligibility, 75% intelligibility) relative to the NSMI group, generally reaching fewer thresholds and at later ages. Children with the added complication of LCI reached fewer thresholds than peers with LCT and did so at even later ages, suggesting that LCI may be a limiting factor for attainment of speech milestones. This finding highlights the complex interactions between speech and language development as well as a potential confounding effect of severity of neurological involvement that may be associated with additional comorbidities (i.e., language and/or cognitive impairment). Mean intelligibility scores at 8 years of age differed for the two groups of children with SMI by nearly 20 percentage points. Children with SMI-LCT had intelligibility at 8 years of 61%; those with SMI-LCI had intelligibility of 42%. Children with SMI showed considerable variability in their intelligibility outcomes, regardless of group membership. The range of variability seemed to be descriptively greater for children in the SMI-LCT group.

Although there were clear differences in speech intelligibility development between the two SMI groups, the age at which children experienced maximum growth did not differ between the groups of children with SMI. Both groups experienced maximum growth between 36 and 60 months of age, with the distribution spread relatively evenly across the 2-year age span for both groups of children (mean age at maximum growth of 46 months for SMI-LCT and 49 months for SMI-LCI). Results from this study for the SMI groups are generally consistent with findings from the earlier companion article where we found that when children were not pooled into profile groups, the age of maximum growth was 36–60 months. Table 6 shows the number of visits with concurrent speech-language therapy by profile group and by age for visits within the 36–60 month window. These results indicate that the majority of children in the SMI groups were receiving speech-language therapy between 36 and 60 months of age. Specifically, there were 141 visits across the two SMI groups from 36 to 59 months of age. Of these, children in the SMI groups were reported to be receiving speech-language therapy for 98 (70%) of visits. We do not have data regarding the specific nature or frequency of speech-language services provided to children. Therefore, we cannot draw any conclusions regarding the effects of therapy. However, it is possible that therapy services may have contributed to rapid change during this critical time for children with SMI. This is an important area for further investigation.

Nearly all children with SMI had considerably diminished speech intelligibility over the course of development, and outcomes at 8 years of age were generally below the 75% intelligibility threshold, regardless of whether or not there was concomitant language comprehension impairment. These findings have important implications for intervention decision making. In particular, augmentative and alternative

Table 6. Number of intelligibility observations by 6-month age band by speech-language profile group.

Age band in months	Profile group	Visits	Visits with concurrent therapy	Age, months *M* (*SD*)	Intelligibility, % *M* (*SD*)	Intelligibility, % Range
24–29	NSMI	2	1	26 (1)	22 (10)	14–29
	SMI-LCT	5	2	27 (3)	27 (17)	9–47
	SMI-LCI	1	1	29	10	10–10
30–35	NSMI	7	3	33 (2)	41 (15)	22–64
	SMI-LCT	13	10	33 (2)	18 (15)	0–41
	SMI-LCI	3	3	33 (2)	19 (4)	15–21
36–41	NSMI	12	6	38 (2)	56 (12)	32–72
	SMI-LCT	20	10	38 (2)	26 (16)	3–53
	SMI-LCI	4	4	39 (1)	23 (7)	14–30
42–47	NSMI	22	6	44 (2)	62 (14)	30–82
	SMI-LCT	27	15	44 (2)	32 (19)	5–65
	SMI-LCI	10	9	45 (2)	28 (21)	0–60
48–53	NSMI	25	5	51 (2)	69 (11)	50–89
	SMI-LCT	29	20	50 (2)	42 (20)	1–73
	SMI-LCI	7	6	51 (2)	38 (20)	16–67
54–59	NSMI	25	6	57 (2)	77 (8)	62–91
	SMI-LCT	31	23	56 (2)	50 (24)	3–82
	SMI-LCI	11	11	56 (2)	27 (19)	0–63
60–65	NSMI	22	4	63 (2)	80 (7)	68–91
	SMI-LCT	30	20	62 (2)	53 (21)	2–83
	SMI-LCI	13	13	63 (2)	33 (22)	3–80
66–71	NSMI	19	1	69 (2)	81 (8)	66–93
	SMI-LCT	28	15	68 (2)	52 (25)	3–88
	SMI-LCI	12	9	69 (2)	34 (23)	0–76
72–77	NSMI	20	1	74 (2)	84 (7)	66–95
	SMI-LCT	27	14	74 (2)	60 (24)	3–88
	SMI-LCI	10	8	74 (1)	29 (16)	4–53
78–83	NSMI	22	2	81 (2)	82 (8)	59–95
	SMI-LCT	29	17	80 (2)	56 (27)	4–89
	SMI-LCI	10	7	80 (2)	35 (17)	11–57
84–89	NSMI	13	1	87 (2)	84 (6)	76–93
	SMI-LCT	17	9	86 (2)	68 (19)	8–91
	SMI-LCI	5	3	87 (1)	34 (18)	11–53
90–96	NSMI	11	0	94 (2)	87 (6)	79–99
	SMI-LCT	17	11	94 (2)	55 (28)	1–89
	SMI-LCI	5	4	95 (2)	39 (23)	9–66
Total	NSMI	200	36			
	SMI-LCT	273	166			
	SMI-LCI	91	78			

Note. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment and typical language comprehension; SMI-LCI = speech motor impairment and language comprehension impairment.

communication (AAC) approaches that serve to enhance speech intelligibility should be considered for all of these children to enhance functional communication and social participation. The specific role that AAC plays for any given children may vary based on the severity of intelligibility deficits, but tools including off-the-shelf technologies (smart phones and tablets) and simple communication boards, used in conjunction with speech or in some cases as an alternative to speech, may provide a powerful means to support communication development in children with CP who have SMI.

Additional Profile Group(s) of Children With SMI Who Show Little Change Over Time

One observation that emerged from this study was that there may be at least one additional profile group of children with CP emerging in our longitudinal data. In our earliest conceptualization of the SLPG paradigm for classifying speech and language abilities in children with CP, we discussed the notion of severity as a differentiator within each of the two profile groups of children with SMI (Hustad et al., 2010). To date, our studies have pooled children across severity levels within each of the two SMI groups (SMI-LCT and SMI-LCI). In this study, there was a set of outliers in each of the SMI groups relating to age of crossing intelligibility thresholds. In particular, in the SMI-LCT group, four children never crossed 25% intelligibility by 100 months (13% of the children in this group). Similarly, in the SMI-LCI group, four children never crossed the 25% threshold by 100 months of age (29% of the children in this group). Some of these children showed change over time in their intelligibility growth, but their predicted maximum intelligibility and growth looked similar, regardless of profile group membership (see Figures 2 and 3). We examined these children individually to determine if there might be common clinical or demographic features. Four of the eight had spastic CP (only one had quadriplegia), one child had ataxic CP, and the type of CP was unknown for three children. One child was Gross Motor Function Classification System Level II, two children were Level III, four children were Level IV, and one child was Level V. Four children were boys; four were girls. We also looked at speech-language therapy history for these children. Results showed that all of the children were receiving therapy services at more than 75% of visits. However, the exact focus of these services is unknown. There did not appear to be a pattern to these characteristics between the two groups. These findings highlight that children with severe SMI and limited speech intelligibility growth over time can be associated with a range of gross motor function levels as well as medical diagnoses. Furthermore, findings support the idea that there may be severity-based subgroups pertaining to speech motor skills among the SLPGs used in this study.

Limitations and Future Directions

There were several key limitations to this study. First, the study is predicated on profile group membership. We assigned profile groups on the basis of speech motor and language skills that were observed prospectively by clinicians with expertise in CP within a research evaluation. Differentiation into profile groups on the basis of SMI was binary in nature and was based on clinical judgment. In reality, speech motor abilities are likely continuous in nature. Future studies could examine children with CP using measurements that permit finer grained characterization of both speech and language abilities. Such studies may suggest additional profile groups or alternative classification paradigms for speech and language abilities.

Speech data examined in this study extended to ages as young as 24 months. However, classifications into profile groups were made at the visit closest to 48 months of age for each child because our earlier work has revealed that many children do not have sufficiently developed speech production abilities to make a determination regarding the presence of speech motor involvement until this time. Thus, our analyses extending down to the youngest ages were looking backward in time at early speech intelligibility given knowledge of later speech and language behavior (and associated profile group membership). This has obvious clinical limitations. However, results such as those provided here provide some insight into how children got to their later classification outcomes. Future studies should seek to address this problem, perhaps with different behavioral measures or different statistical approaches.

In our earlier companion article (Hustad, Sakash, Natzke, et al., 2019), we noted that a major limitation of this work is the lack of large-scale normative data on typically developing children against which to compare children with CP. This limitation is further magnified in the present study with the finding that children with NSMI show different outcomes and different intelligibility growth patterns than either of the groups of children with SMI. Normative data that would allow us to compare these children with typically developing peers could further our understanding of the impact of CP on speech development, even for those without frank speech motor involvement. It would also help establish standards for identification of what is “normal” for functional intelligibility.

Intelligibility is a complex construct that can be measured in different ways. In this study, naïve listeners orthographically transcribed the speech of children. Two listeners heard each child, and an average intelligibility score across the two listeners for each child was examined as our dependent measure. Listeners can vary considerably in their performance on intelligibility tasks, even when hearing the same speech samples. The ideal or even a sufficient number of listeners necessary to represent the range of variability across listeners, yielding a stable mean intelligibility score for any given speaker, has not been established in the literature. However, future studies should examine the stability of findings such as those of this study when more than two listeners hear each child.

Single-word intelligibility has inherent limitations given that children generally speak in utterances that are longer than one word in length. Studies of connected speech are needed to examine the impacts of profile group membership on more ecologically valid speech samples, including production of multiword utterances and conversational speech. In addition, our intelligibility data were obtained in a highly controlled laboratory environment using standard orthographic transcription procedures. Studies examining other ways to measure intelligibility (such as rating scales) may be useful for establishing metrics that are more readily adaptable to clinical settings where it may not be feasible to obtain intelligibility ratings using unfamiliar listeners.

Clinical Implications

This study has several potential clinical implications that affect treatment decision making for children with CP. Results of this study revealed clear evidence that classifying children with CP on the basis of SLPGs (following Hustad et al., 2010) yields important differences among groups with regard to the age of crossing intelligibility thresholds, intelligibility growth, attainment of intelligibility outcomes, and prediction of intelligibility outcomes at 8 years on the basis of data from 3 years of age. Our results highlight that children with CP who have NSMI have substantially different growth and better intelligibility outcomes than their peers who have SMI. Most of these children are not clear candidates for therapy focused on intelligibility at present; however, beyond a small preliminary data set (Hodge & Gotzke, 2014), we do not currently have parallel measures on typically developing children, so we cannot definitively quantify the extent to which children with NSMI have intelligibility development that is consistent with age expectations. It is possible that these children may benefit from therapy focused on other speech language issues. Studies are needed to quantify the speech and language abilities of children with CP and NSMI to determine the extent to which other speech and language challenges beyond SMI may be present.

Children with SMI with or without comorbid language comprehension impairment have considerable differences in speech intelligibility relative to children with NSMI in terms of rate of intelligibility growth, timing of growth, age of acquisition of different intelligibility thresholds, and outcomes at 8 years. All children in both SMI groups would be candidates for speech-language therapy based on the presence of dysarthria; most of the children in the SMI groups had significantly reduced intelligibility and delayed intelligibility development, and many never reached speech intelligibility levels that might be considered clinically "adequate" for communication. Children who have SMI-LCT tend to have better outcomes than children with SMI-LCI, although there was considerable variability among children. Early intensive intervention is important for all children with SMI; however, it may be especially important for children with comorbid language challenges who generally have poorer intelligibility outcomes than children with SMI-LCT. AAC interventions should be considered for all children with SMI to support speech development and to foster functional communication and social participation.

There may be a subset of children with SMI, across both language groups, who fall outside the range of the growth and intelligibility outcome profiles for the larger groups (i.e., SMI-LCT and SMI-LCI). These children may comprise a separate subgroup that is characterized by very low intelligibility throughout the course of development with relatively minimal change over time. These children clearly require special attention relative to the other profile groups because of the severity of their speech involvement. Specifically, these children require comprehensive AAC systems to support communication.

Finally, in our earlier analysis, we showed that intelligibility at 36 months of age was highly predictive of intelligibility at 8 years of age. In this study, we showed that this prediction could be improved by a small amount based on knowledge of a child's profile group. However, classifying children into profile groups is often difficult prior to the age of 4 years because speech in children with CP is frequently delayed in its onset (Hustad et al., 2017) and features of dysarthria are difficult to differentiate from early speech development features in young children. Therefore, we suggest that when considering prediction of outcomes at 8 years based on 36-month data, our earlier results may be the most ecologically valid. However, in considering intelligibility growth, rate of change, age of crossing intelligibility thresholds, and achievement at 8 years, data based on profile groups as presented in this study provide a more refined perspective on children with CP.

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