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Research Article

Longitudinal Growth in Single Word Intelligibility Among Children With Cerebral Palsy From 24 to 96 Months of Age: Predicting Later Outcomes From Early Speech Production

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Purpose: Children with cerebral palsy (CP) are at risk for significant communication problems. Reduced speech intelligibility is common, even for those who do not have speech motor deficits. Development of intelligibility has not been comprehensively quantified in children with CP; as a result, we are currently unable to predict later speech outcomes. Such information would advance treatment decision making. We sought to examine growth in speech intelligibility among children with CP using a prospective longitudinal design, with a focus on age of crossing target intelligibility thresholds, age of greatest intelligibility growth, and how well intelligibility at 36 months predicted intelligibility at 96 months.
Method: Sixty-nine children with CP were followed longitudinally between 24 and 96 months of age. A total of 566 time points were examined across children (M = 8.2 time points per child, SD = 2.6). We fitted a nonlinear random effects model for longitudinal observations and then used the fitted model trajectories to generate descriptive analyses of growth. We used results of the model to generate a set of simulations, which we analyzed to determine how well 36-month intelligibility data predicted 96-month data.
Results: Half of children crossed 25% and 50% intelligibility thresholds at 36 and 49 months of age, respectively. Slightly more than half of children did not reach 75% intelligibility by 96 months of age. Age of crossing 25%, 50%, and 75% intelligibility thresholds was highly negatively correlated with intelligibly at 96 months. Children had the steepest intelligibility growth at 36 months, followed by 48 and 60 months. Intelligibility at 36 months was highly predictive of intelligibility at 96 months.
Conclusions: The developmental window from 3 to 5 years constitutes a time of rapid growth in speech intelligibility in children with CP. Children who cross intelligibility thresholds of 25%, 50%, and 75% at earlier ages have better outcomes when they are older; early performance is highly predictive of later speech intelligibility outcomes. Children with CP as a group have delayed speech intelligibility development but are still growing through 96 months of age.

Children acquire the ability to produce intelligible speech through a complex and protracted developmental process involving, at a minimum, speech motor, sensory, linguistic, and cognitive domains. The estimated 2.5 per 1,000 children who have cerebral palsy (CP; Paneth, Hong, & Korzeniewski, 2006) are at significant risk for a variety communication disorders because of potential deficits affecting speech motor control, cognitive, linguistic, and sensory abilities. One study found that 60% of children with CP had some type of communication difficulty based on the impressions of physicians (Bax, Tydeman, & Flodmark, 2006). Studies examining speech and language development in children with CP using finer grained measures and methods have found that, in younger children with CP, speech and language delays may be quite common (Hustad, Allison, McFadd, & Riehle, 2014). Different speech-language profile groups have been documented in children with CP (Hustad et al., 2014; Hustad,

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Gorton, & Lee, 2010; Hustad, Oakes, McFadd, & Allison, 2016). Variations in these profiles include the presence or absence of speech motor involvement and the presence or absence of language/cognitive involvement. The speech motor disorder *dysarthria* is estimated to occur in about 50% of the population (Nordberg, Miniscalco, Lohmander, & Himmelmann, 2013) and is reflected in several of the aforementioned speech-language profile groups. Dysarthria can affect any one or multiple speech subsystems, but studies suggest that the articulatory subsystem is a key contributor to speech motor deficits observed in children with CP (Allison & Hustad, 2018; Chen, Hustad, Kent, & Lin, 2018; Lee, Hustad, & Weismer, 2014; Nip, 2015). A hallmark feature of dysarthria is reduced speech intelligibility (Darley, Aronson, & Brown, 1969). Recent studies suggest that even children who have CP with no clinical signs of dysarthria have reduced intelligibility relative to typically developing peers (Hustad, Sakash, Broman, & Rathouz, 2018; Hustad, Schueler, Schultz, & DuHadway, 2012).

Intelligibility has been defined as the extent to which an acoustic signal, generated by a speaker, can be correctly recovered by a listener (Kent et al., 1989; Yorkston & Beukelman, 1980). Intelligibility is a dyadic construct, and its measurement reflects the joint efforts of the speaker (who produces the signal) and the listener (who interprets the signal; Lindblom, 1990). To be intelligible, speech does not need to be perfect or even "normal." The key issue is whether listeners are able to map the acoustic signal onto the intended lexical units. A considerable body of research has demonstrated that there are many variables that may influence intelligibility. These variables include, but are not limited to, age (Hodge & Gotzke, 2014a), utterance length and complexity (Allison & Hustad, 2014), listener familiarity with the speaker and experience with listening to child or dysarthric speech (D'Innocenzo, Tjaden, & Greenman, 2006; Liss, Spitzer, Caviness, & Adler, 2002), and quantitative characteristics of the speech signal, including temporal and spectral production features (Allison & Hustad, 2018).

In the study of speech motor disorders, intelligibility is generally measured either objectively or subjectively. Subjective measures require listeners to quantify their perception of a speaker's intelligibility by assigning a number to or scaling what they heard (Darley et al., 1969; Platt, Andrews, Young, & Quinn, 1980; Weismer & Laures, 2002). Objective measures involve transcription (usually orthographic) or forced-choice recognition of words by listeners, typically yielding a percentage of words identified correctly relative to the target words that the speaker intended to produce (Tikofsky & Tikofsky, 1964; Yorkston & Beukelman, 1978, 1980). An advantage to this approach is that quantification is straightforward: Lexical units are either correct or incorrect. In order to score lexical units, however, target words produced by the speaker must be known so that they can be scored accordingly. For this reason, elicited words and sentences are typically used for measuring intelligibility via transcription or forced-choice recognition approaches.

Studies of intelligibility and its development in children, both disordered and typical, are limited. Only a small number of studies have attempted to quantify intelligibility of words (either in isolation or in connected speech) in typical children, and there are currently no comprehensive quantitative data that document intelligibility expectations across the age span using a single consistent methodology. Existing studies examining children have primarily employed listeners who are "experts" (commonly speech-language pathologists 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 naïve listeners (Baudonck, Buekers, Gillebert, & VanLierde, 2009) and that learning occurs for listeners over time (Hustad, Oakes, & Allison, 2015). Only a few studies (see Hodge & Gotzke, 2014a, 2014b) have used naïve listeners to objectively characterize intelligibility. A more complete understanding of intelligibility development in typical and disordered populations is critical toward establishing benchmarks and cut-points for development. However, it is essential that this be done within a framework that has clinical relevance for children with speech motor disorders, such as those with CP. The study of functional intelligibility growth would begin to address this need.

Regardless of how it is quantified, reduced intelligibility has significant negative consequences for functional communication, social participation, and quality of life (Dickinson et al., 2007; Schliephake, Schmelzeisen, Schönweiler, Schneller, & Altenbernd, 1998). Enhancing intelligibility is often a primary goal of intervention (Ansel & Kent, 1992), and thus, it has received considerable attention in the adult dysarthria literature (Fletcher, McAuliffe, Lansford, Sinex, & Liss, 2017; McAuliffe, Fletcher, Kerr, O'Beirne, & Anderson, 2017; Stipancic, Tjaden, & Wilding, 2016) and in a growing body of pediatric dysarthria literature (Allison & Hustad, 2014; Hustad et al., 2012; Pennington, Lombardo, Steen, & Miller, 2018; Pennington, Miller, Robson, & Steen, 2010). Important clinical decisions are often made from speech intelligibility data. For example, intelligibility scores serve as a basis of comparison for documenting and monitoring change in speech performance (Yorkston, Beukelman, Strand, & Bell, 1999), as a measure of severity of the speech disorder (Weismer & Martin, 1992), and as an index of functional ability (or disability) relative to normal performance (Yorkston et al., 1999).

Heterogeneity among children with CP makes the study of speech intelligibility and its development difficult. Because cross-sectional data involving different children do not provide an accurate picture of change over time, prospective longitudinal methods where each child is his or her own control are necessary for the study of development in children with CP. Longitudinal methods allow for direct connection between early speech-language markers and later development as well as characterization of the range of longitudinal trajectories in development. Results from this type of study are critical to inform our understanding of rates and limits of developmental change so that we can begin to predict outcomes and use this information to develop interventions that change growth trajectories. The ability to predict later outcomes, particularly related to speech intelligibility, is also useful for identifying the need for augmentative and alternative communication (AAC) systems and strategies to support speech and overall communication development at very early ages, rather than using a "wait and see" approach with speech development. For example, if we can predict, based on data obtained at 2 or 3 years of age, that a child's intelligibility at 8 years of age will be only 70%, this would provide clear and convincing evidence that AAC interventions should be implemented as soon as possible to ensure that children have access to tools and strategies to support expressive communication at the earliest possible age.

Longitudinal work has yielded important and clinically useful findings to date. For example, one study examined how well the age at which children with CP became able to produce single words in an elicitation task predicted intelligibility and maximum length of utterance at 53 months (Hustad et al., 2017). Findings showed that most children, including those who had speech motor impairment and those who did not, had intelligibility of about 20% when they first became able to produce elicited words, regardless of their age, and that they made dramatic gains in word intelligibility immediately after becoming able to produce elicited words. Similar rapid gains would be expected for typically developing children, but parallel research has not been conducted. It is noteworthy, however, that, in children with CP, the age at which children began producing elicited words had a significant impact on the rate of change in intelligibility and utterance length, such that children who produced single words in imitation at earlier ages tended to make faster gains in intelligibility and faster gains in length of utterance, ending with higher intelligibility and longer utterances at 53 months. In the same study, Hustad et al. (2017) found that children with CP who had the best outcomes were able to produce elicited words by 24 months of age. However, even the most advanced children, as a group, had reduced intelligibility relative to what would be expected for typically developing peers (Hustad et al., 2012), indicating a clear need for intelligibility-related intervention throughout the preschool years.

We sought to extend our earlier longitudinal work, which examined a small group of children with CP up to 53 months of age, to a larger group of children across a wider age range in order to begin to understand how speech intelligibility develops in children with CP. Growth curves, similar to what we sought to develop in this study, have been created for a number of different domains, particularly gross motor development (Palisano et al., 2000; Rosenbaum, Palisano, Bartlett, Galuppi, & Russell, 2008; Rosenbaum et al., 2002; Wood & Rosenbaum, 2000). Information from growth curves has been useful for describing population characteristics and for predicting later motor abilities. Similar research in the speech domain is essential to understand the course of development of speech in children with CP and to advance the empirical basis for treatment decision making.

In this study, we examine longitudinal growth of single word intelligibility. Single word intelligibility differs from connected speech intelligibility in many important ways, including the presence of linguistic context, and coarticulatory blending of acoustic information across word boundaries. Studies of adults have repeatedly demonstrated that intelligibility scores tend to be lower for single word stimuli than for sentences (Miller, Heise, & Lichten, 1951; O'Neill, 1957; Salasoo & Pisoni, 1985; Sitler, Schiavetti, & Metz, 1983; Yorkston & Beukelman, 1981) and narratives (Hustad, 2008). Thus, examining single word intelligibility provides a starting point, but not a complete picture of intelligibility in children with CP. In addition, use of single words allows for the inclusion of children with CP who have limited speech production abilities, as well as those with advanced speech production abilities, allowing characterization of the full range of developmental profiles among children with CP and quantification of how children change over time. We pursued such an approach in this longitudinal study of a cohort of children with CP. Our longitudinal methodology allowed us to examine each individual child with CP relative to himself or herself, to look at trajectories of change across many children, and to characterize and quantify the degree to which early childhood measures of intelligibility predict later childhood outcomes, that is, the degree to which children track over time.

We focused on children with CP between the ages of 24 and 96 months because we expect production of speech to be established by 24 months in typical children and we know that typical children are continuing to show refinement in their speech production abilities, as indicated by segmental articulation development norms, through 96 months of age. Thus, we expected to be able to capture important developmental change in our target age range for children with CP. We used a consistent corpus of words that were produced at each longitudinal visit from children to examine how children change over time on production of the same stimuli. We also used an orthographic transcription paradigm with unfamiliar listeners to reduce the potential of listener learning as a contributor to change in intelligibility. This would provide an unbiased picture of change over time in listener's ability to decode the speech signal and map it onto words. This type of measurement is consistent with longstanding approaches to characterizing intelligibility of dysarthric speech, yielding functional measures of single word production. Importantly, we sought to use this information to describe variability in age points for crossing different clinically relevant intelligibility thresholds, to characterize variability in children's trajectories and outcomes, to identify periods of maximum growth that could be leveraged for intervention, and to predict later outcomes based on earlier data. We addressed the following specific research questions:

1. What is the distribution of ages at which children reach 25%, 50%, and 75% intelligibility thresholds for single words?

2. How intelligible are children with CP at 96 months of age? How much heterogeneity across children is there in this end point?
3. What is the relationship of intelligibility at 96 months to age of crossing 25%, 50%, and 75% thresholds for single words?
4. At what ages do children have the greatest growth in single word intelligibility?
5. How well does single word intelligibility at 36 months predict single word intelligibility at 96 months?

Because this study was the first of its kind to prospectively examine the longitudinal development of intelligibility in children with CP, we did not have a priori hypotheses regarding our research questions beyond the general expectations that intelligibility would improve with age in all children and that intelligibility at early ages would, to some degree, predict intelligibility at later ages. Instead, we sought to characterize the natural course of development among children with CP as they grew over time. We selected threshold points at 25%, 50%, and 75% intelligibility because they have potential clinical relevance with regard to functional interpretation and there is precedent from a widely cited parent report study of intelligibility development in typical children (Coplan & Gleason, 1988). In subsequent work, we will examine the data presented in this article in different ways that separate children in a more specific and fine-grained manner to further refine our understanding of intelligibility development based on features of the underlying pathology and impairment. In this article, however, our aim was to provide a descriptive account of what intelligibility development looks like in children with known risk for dysarthria and to characterize how early results can predict later outcomes.

Method

Participants

Sixty-nine children with CP (33 girls, 36 boys) participated in this study. Children were a subset of those participating in a larger longitudinal study of communication development in CP. For the larger study, children were between 18 and 60 months at initial enrollment, had a medical diagnosis of CP, and had hearing within normal limits as documented by either formal audiologic evaluation or distortion product otoacoustic emission screening. Inclusion in this study required that children (a) were able to produce speech, which was operationally defined as the ability to repeat single words in an elicitation task; (b) had contributed at least two longitudinal time points to the larger study in which they produced speech; and (c) had no codiagnosis of autism spectrum disorder. Data were collected from children twice yearly at 6-month intervals until their eighth birthday, after which they were seen for yearly visits. Across the 69 children who met inclusion criteria, each child contributed two to 13 data points, for a total of 566 data points, yielding a mean of 8.2 (SD = 2.6) and a median of 9 data points per participant. All children were from homes where American English was the primary language. Children were born in the United States between 2000 and 2009. Demographic information including CP diagnosis is presented in Table 1.

During the time frame of this study, children were receiving whatever therapy was provided to them in their usual environment. We gathered information via parent report regarding whether or not children were currently enrolled in speech-language therapy at the time of each visit. Findings regarding enrollment in therapy are provided in Table 2. Generally, about half of children were receiving speech-language therapy at any given age point, with some fluctuation over time. Note, however, that the specific nature and frequency of therapies provided and the progress of individual children in therapy are not known.

There were also 1,132 nondisabled adult listeners who participated in this study. Two different listeners were quasi-randomly assigned to each child and each visit (566 visits across the 69 different children × 2 listeners per visit = 1,132 listeners); each listener heard only one child at one visit producing all stimulus material. Listeners were recruited from the university community via public postings and were primarily undergraduate students. Listeners were compensated monetarily for their participation. Inclusion criteria required that listeners (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) be between 18 and 45 years of age; (c) have no more than incidental experience listening to or communicating with persons having communication disorders; (d) be a native speaker of American English; and (e) have no identified language, learning, or cognitive disabilities per self-report. Listeners were 305 males and 827 females. The mean age of listeners was 20.8 years (SD = 5.6).

Materials and Procedures

Children participated in a standard speech and language assessment protocol for each visit. The protocol was administered by a research speech-language pathologist in a sound-attenuating suite. For this study, speech production—and in particular speech intelligibility—results were the primary focus.

Acquisition of Speech Samples From Children

For each visit, children produced a corpus of speech stimuli from the Test of Children's Speech (TOCS+; Hodge & Daniels, 2007). Stimuli included single word productions that were the same for each visit and for each child. TOCS+ stimuli were developed to be appropriate (lexically, phonetically, syntactically, and morphologically) for children and have been used regularly in related research (Hodge & Gotzke, 2014a, 2014b). For this study, we focused on single word productions, which comprised 38 different words from the TOCS+. Words included all items from the TOCS+ 30-word probe (Hodge & Daniels, 2007), as well as eight additional words, which were added to ensure adequate representation of

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

Characteristic	Children with CP (*N* = 69)
Mean number of visits (*SD*)	8.2 (2.6)
Median number of visits	9
Male/female ratio	36:33
Type of CP	
Spastic	
Diplegia	14
Hemiplegia	27
Triplegia	2
Quadriplegia	8
Dyskinetic	1
Ataxic	5
Mixed	2
Unknown	8
GMFCS at 54 months	
I	43
II	6
III	8
IV	9
V	3
Speech-language profile group at 54 months	
No speech motor involvement	23
Speech motor involvement–typical language	26
Speech motor involvement–impaired language	20
Anarthric	0

Note. GMFCS = Gross Motor Function Classification System Rating (Palisano et al., 1997).

corner vowels. In total, the TOCS+ words sampled 74 consonant targets (19 unique consonant types) and 44 vowel targets (15 unique vowel types), for a total of 35 unique phoneme targets. In addition, there were seven unique syllable shape types. Lexical characteristics of words such as word frequency, neighborhood density, and phonotactic probability of TOCS+ words were not controlled.

Speech samples were obtained using elicitation procedures in which children were presented with adult recordings of each target word, along with an image depicting the word via a portable screen. Children then repeated what they heard following the recorded model. Productions were monitored online by a research assistant to ensure that samples were free of overlap with examiner speech and free of extraneous noises. Children were asked to repeat productions when these criteria were not met. Having children produce a known corpus of words that is the same across all children allowed us to compare listener orthographic transcriptions against known target responses, thus ensuring that intelligibility scores were an accurate reflection of which target words were perceived correctly by listeners.

Speech samples from children were recorded using a digital audio recorder (Marantz PMD 570) at a 44.1-kHz

Table 2. Speech-language therapy services at the time of each visit per parent report (*N* = 69 children).

Pattern of therapy services over time	Number of children	Mean number of visits where therapy reported (*SD*)	Median number of visits where therapy reported
Children who never received speech-language therapy over the course of participation	16	NA	NA
Children who received speech-language therapy for the entire course of participation	14	7.28 (2.89)	8
Children who received a period of therapy, which was discontinued during the course of participation	20	5.25 (2.39)	6
Children who received intermittent therapy over the course of the study	19	3.89 (2.07)	6
Number of children who received AAC intervention at any point over the course of the study	12	.69 (1.62)	0

Note. Therapy information was not provided for 10 visits across a total of six children. AAC = augmentative and alternative communication.

sampling rate (16-bit quantization). A condenser studio microphone (Audio-Technica AT4040) was positioned next to each child using a floor stand and was located approximately 18 in. from the child's mouth. The level of the signal was monitored and adjusted on a mixer (Mackie 1202 VLZ) to obtain optimized recordings and to avoid peak clipping.

Acquisition of Intelligibility Data

Digital recordings of children's speech were edited on a personal computer to remove extraneous noises and the speech-language pathologist's voice. We created individual .wav files for each stimulus utterance and peak amplitude–normalized each to assure that the maximum loudness levels of the recorded speech samples were the same across children and individual words, while preserving the amplitude contours of the original productions. Files were then played back to listeners.

Speech stimuli were delivered to listeners via in-house software that presented audio samples in a self-paced experimental task and stored the resultant-typed orthographic transcriptions. During the listening task, listeners were seated individually in a sound-attenuating suite in front of a 19-in. flat-panel screen with a keyboard placed directly in front of them. An external speaker was connected to a computer and situated directly beneath the computer screen. The peak audio output level was calibrated to approximately 75 dB SPL from where listeners were seated and was checked periodically to ensure that all listeners heard stimuli at the same output level. Individual words produced by children were randomized for each listener so that no two listeners heard the same words in the same sequence. Individual words were presented to listeners in isolation without a carrier phrase. Listeners were allowed to hear each production one time and were told that the purpose of the study was to determine how understandable children were to unfamiliar listeners like themselves. 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. Listeners were provided with instructions on how to use the experimental software to advance through the experiment and type in their orthographic response for each word presented. In addition, they heard four sample productions to familiarize themselves with the experimental task. Data from the sample productions were excluded from analyses. Data collection from listeners took approximately 30 min per listener.

Analysis of Speech Intelligibility Data

Typed orthographic transcriptions of each stimulus word produced by each child were generated by two independent listeners per child per visit. Listeners' typed orthographic transcriptions of children's word productions were converted to phonetic transcriptions using in-house customized software. Phonetic transcriptions of listeners' responses were compared against phonetic transcriptions of the word target produced by children. Note that these were transcriptions of the target words children were trying to produce and not narrow phonetic transcriptions of the actual productions from children. This is a standard approach in speech motor disorders research (Hodge & Gotzke, 2014a, 2014b; Yorkston & Beukelman, 1978, 1980), and it allowed us to consider intelligibility from a lexical perspective, which was the goal of the study. Intelligibility scores for each child and each visit were obtained by counting the number of words transcribed correctly by each listener relative to the target words that children were attempting to produce. Listener transcriptions that were an exact phonemic match to the target word were counted as correct. Misspellings and homonyms were accepted as correct, as long as all phonemes in the listener transcription matched the target words (e.g., if a listener typed "there" and the target word was "their," the listener's target was counted as correct). 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.

Statistical Analyses

Analyses of the longitudinal intelligibility data with the goal of describing and quantifying the between-children heterogeneity of intelligibility trajectories required management of several challenging features: First, the age range of visits varied somewhat from child to child. Second, the number of visits was also variable across children, and owing to intermittent missingness, the visits were irregularly spaced for some children. Third, growth in intelligibility as a function of age is highly nonlinear, eventually flattening out or plateauing at later ages. For these three reasons, it was challenging to fit an individual growth curve separately to each child's data. Fourth, even while generally following a smooth trajectory of growth, the within-child variability of measures around that trajectory was substantial. To address these concerns, we fitted a nonlinear random effects model for longitudinal observations to the data, with random effects at the child level (Davidian, 2017). In this approach, the fixed effects part of the model captures an average trajectory, whereas the random effects at the child level quantify deviations for each child from that average trajectory, resulting in an estimated individual trajectory for each child. We then based our descriptive analyses of the data on the fitted model for each child. The model resolves the aforementioned issues by objectively smoothing out the data for each subject and by borrowing information across subjects when data for an individual subject are sparse, while letting the individual data largely "speak for itself" for children who have denser coverage of visits. It provides best predictions for subject-level parameters, which are only imperfectly quantified by the observed data. As such, the approach captures the individual variability by striking a balance between using each child's data in isolation—which would overstate the variability from child to child—and assuming that all children follow a single common trajectory, which would obviously understate the heterogeneity. For example, the fitted model will provide, for each subject, best predictions of the maximum attained intelligibility value (Research Question 2) or of the age at maximum growth in intelligibility (Question 4). Our research questions are addressed through descriptive analyses of the distributions of these predictions.

To begin our analyses, we plotted intelligibility measurements against age in months for each child. The nature of the intelligibility score as a proportion from 0 to 1 led us to model this score against age as a modified logistic function with normally distributed residuals, with growth bounded by an upper limit, separately estimated for each child. We chose the model family based on the characteristics of the response measures and used mixed-effects models that fully exploited the association among measurements from the same participant.

The parameters estimated for the logistic function (Equation 1) are the asymptote (*Asym*) or the maximum/plateau value on the curve, the value of time t at the inflection point of the curve (*tmid*), and a numeric scale parameter representing the rate of change (*scal*). The realized measure at time t is assumed to be normally distributed with mean $f(t)$.

$$f(t) = \frac{Asym}{\left(1 + e^{\left(\frac{tmid - t}{scal}\right)}\right)} \quad (1)$$

Equation 1. Logistic function with parameters *Asym* = asymptote, *xmid* = inflection point, and *scal* = rate of change.

Importantly, each participant was accorded random effects both in the asymptote and in the midpoint, allowing considerable between-subjects variability in trajectories, including the plateau intelligibility (*Asym*) that the child will ultimately reach. We used the fitted models—including the predicted values of the random effects for each subject—to predict ages at which specified intelligibility thresholds are crossed. We captured these in box plots and dot plots in the margins of the scatter plot distributions.

The fifth research question addressed how well single word intelligibility at an early age (36 months) would predict single word intelligibility at a later age (96 months). The nonlinear mixed-effects model in Equation 1 provided a predicted trajectory of intelligibility for each subject and provided estimates of the distribution of these trajectories (captured by the variance and covariance of the random midpoint and the random asymptote). We sampled with replacement from these subjects and, using each subject's parameter and covariance estimates, simulated new parameters from a multivariate normal distribution to generate 1,000,000 trajectories. We removed trajectories with simulated intelligibility above 100 or below 0. From each of these trajectories, we pulled intelligibility points from 36 and 96 months. We then used a nonlinear regression model of simulated 96-month intelligibility versus simulated 36-month intelligibility to depict and quantify the relationship. Returning to the original data, observed intelligibility collected within a 12-month window of 36 and 96 months was compared with the simulated data set as a check on our modeling approach.

Results

Descriptive results for observed data from children with CP are shown in Figure 1 and Table 2. Summary statistics suggest that children showed a steady increase in intelligibility over time, particularly in the earlier years. These data also suggest that there is considerable variability within each age band, which is not unexpected given that the sample included a full range of children with CP who were able to produce single word utterances, including those with dysarthria and those without dysarthria.

Question 1: What is the distribution of ages at which children reach 25%, 50%, and 75% intelligibility thresholds for single words?

Figure 1 shows observed trajectories of each of the 69 children who contributed two or more longitudinal data points to the data set. Box plots with multicolored dot plots below the trajectory plot are distributions of model-predicted age points at which a child's trajectory would cross 25%, 50%, and 75% intelligibility thresholds. Model-predicted ages greater than 100 months are represented on the far right of each dot plot, indicating that children's trajectories did not reach the target intelligibility by 100 months of age. Histograms in the upper three panels of Figure 3 display the percentage of children by age reaching each of the three target threshold points.

Descriptive results from the model depicted in Figures 2 and 3 suggest that 25% of children with CP have reached the 25% intelligibility threshold by 29 months of age, 50% have reached the 25% intelligibility threshold by 36 months of age, and 75% of children have reached the 25% intelligibility threshold by 49 months of age. The modeled data suggest that 12% of children do not reach 25% intelligibility by 100 months.

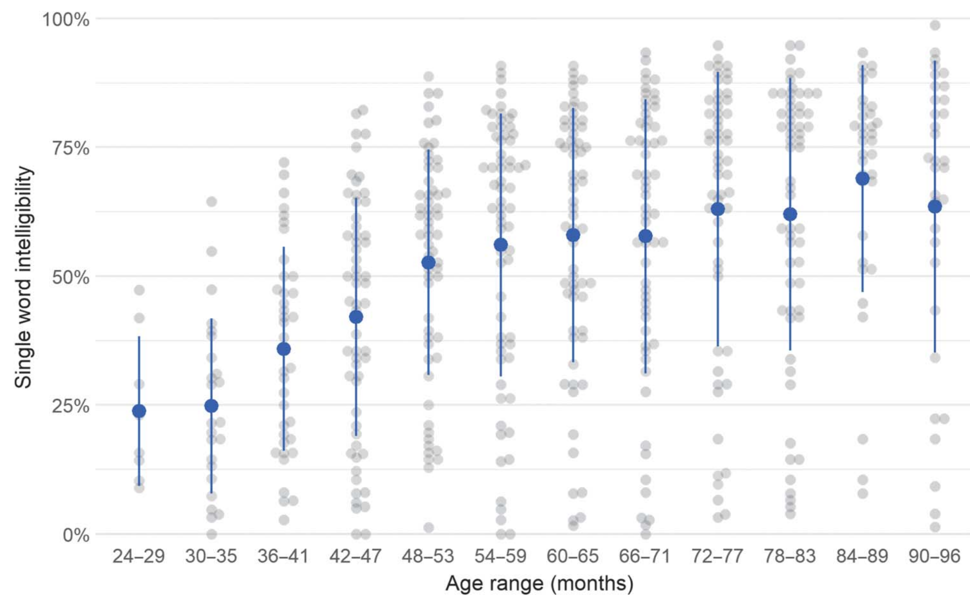
For the 50% intelligibility threshold, results from the model suggest that 25%, 50%, and 75% of children have reached the threshold by 40, 49, and 84 months of age, respectively. Modeled data indicate that 23% of children do not reach the 50% intelligibility threshold by 100 months.

Finally, for the 75% intelligibility threshold, results from the model suggest that 25% of children with CP have reached the threshold by 58 months of age. However, modeled data suggest that (at least up to the 100-month age range modeled here) relatively few additional children (only 20%) reach 75% intelligibility after 60 months and 38 children in the sample (56%) do not reach the 75% intelligibility threshold by 100 months.

Question 2: How intelligible are children with CP at 96 months of age? How much heterogeneity across children is there in this end point?

The vertical axis in Figure 2 shows a box plot/dot plot of model-predicted maximum intelligibility scores attained up to 96 months of age. Descriptive results from the model show that the 50th percentile (median) intelligibility was 73% at 96 months, indicating that half of children had intelligibility above 73% at 96 months and half had intelligibility below this. Results from the model also indicate that

Figure 1. Observed single word intelligibility scores by 6-month age band for children with CP. Individual data are plotted in gray, blue dots represent means, and upper and lower error bars show standard deviations.



the lower and upper quartiles, respectively, were 51% and 82% at 96 months of age, suggesting substantial variability in the 96-month end point for single word intelligibility among children with CP.

Question 3: What is relationship of intelligibility at 96 months to age of crossing 25%, 50%, 75% thresholds for single words?

Scatter plots in the lower three panels of Figure 3 show the relationship of maximum intelligibility to age of reaching three different target intelligibility thresholds. The relationship of predicted maximum intelligibility to age of reaching 25% intelligibility indicates a very strong negative correlation of $r = -.83$ ($n = 61$ children), such that children who reach 25% intelligibility at younger ages have higher maximum single word intelligibility at later ages. Note that the eight children not included in this correlation never reached the 25% intelligibility threshold and are not included in the correlation calculation.

The relationships of maximum intelligibility to age of reaching 50% and 75% intelligibility are similarly strong, with $r = -.84$ ($n = 53$) and $r = -.72$ ($n = 31$), respectively. Note that 16 and 34 children are not included in these calculations because they never achieve 50% or 75% intelligibility.

Collectively, these results suggest that children who reach intelligibility thresholds of 25%, 50%, and 75% at earlier ages have better speech outcomes later in life than those whose intelligibility development is more delayed. In addition, more than half of children in this sample never reach 75% intelligibility for single word productions.

Question 4: What are the ages when children have the greatest growth in single word intelligibility?

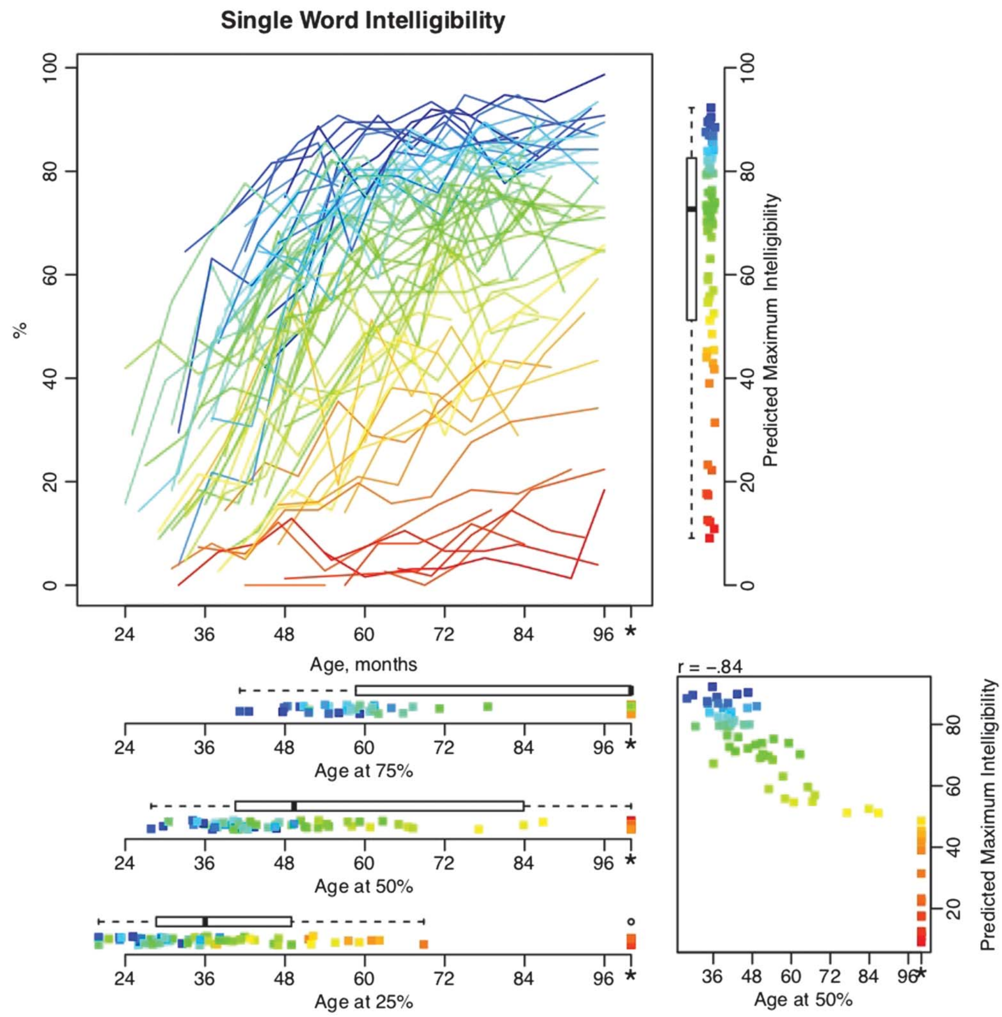
The statistical model we used to generate descriptive statistics assumes a sigmoidal S-curve tracing out, for each child, increasing intelligibility across age. As such, each curve admits a point of maximum growth where the curve is the steepest with respect to age. Figure 4 shows a histogram of the distribution of ages (represented as percentage of the sample on the vertical axis) at which children have the steepest (maximum) growth in their modeled age trajectory of intelligibility. The greatest proportion (45%) of the sample shows the steepest intelligibility growth around 36 months of age, followed by steepest growth around 48 months of age (27% of the sample) and around 60 months of age (17% of the sample).

The scatter plot in Figure 4 shows the relationship between age of steepest intelligibility growth and maximum predicted intelligibility score. The correlation between these two variables is $r = -.83$, indicating that children who have their steepest growth at younger ages have the best intelligibility outcomes at the age of 8 years.

Question 5: How well does single word intelligibility at 36 months predict single word intelligibility at 96 months?

Figure 5 shows a plot of simulated single word intelligibility at 96 months against simulated intelligibility at 36 months; the simulated data are based on the fitted statistical model used for the analyses addressing Questions 1–4. Overall, intelligibility scores at 36 months predict intelligibility scores at 96 months with an R^2 value of .78 for the curvilinear relationship of 96-month intelligibility to 36-month intelligibility. However, examination of Figure 5 suggests that intelligibility scores below 10% at 36 months have little predictive value: For these scores, intelligibility at 96 months varied widely, ranging from 0% to as high as 80%. The highest predictive ability appears to be for scores between 10% and 40% at 36 months, where there is a strong association of lower scores at 36 months and lower

Figure 2. Observed trajectories plus the age crossing for 25%, 50%, and 75% single word intelligibility thresholds. Box plots below the trajectory plot are a distribution of predicted ages at which a subject's trajectory would cross the *X*% intelligibility threshold, where *X* = 25%, 50%, and 75% intelligibility. Predicted ages that fall outside the 20- to 100-month range are plotted at the extremes of these ranges. Subject colors are ordered by their predicted maximum intelligibility for the first random effect parameter, the random asymptote.



scores at 96 months. Above about 40% intelligibility at 36 months, the relationship of 96- to 36-month intelligibility was somewhat flatter.

Discussion

The purpose of this study was to provide an initial descriptive account of speech intelligibility development in a diverse set of children with a diagnosis of CP. Our goal was to describe speech intelligibility development trajectories across a range of children with CP, regardless of whether dysarthria was present. Key findings from this study were as follows.

First, the developmental window from 3 to 5 years of age seems to be an important time for speech intelligibility development, wherein children with CP are growing the most rapidly.

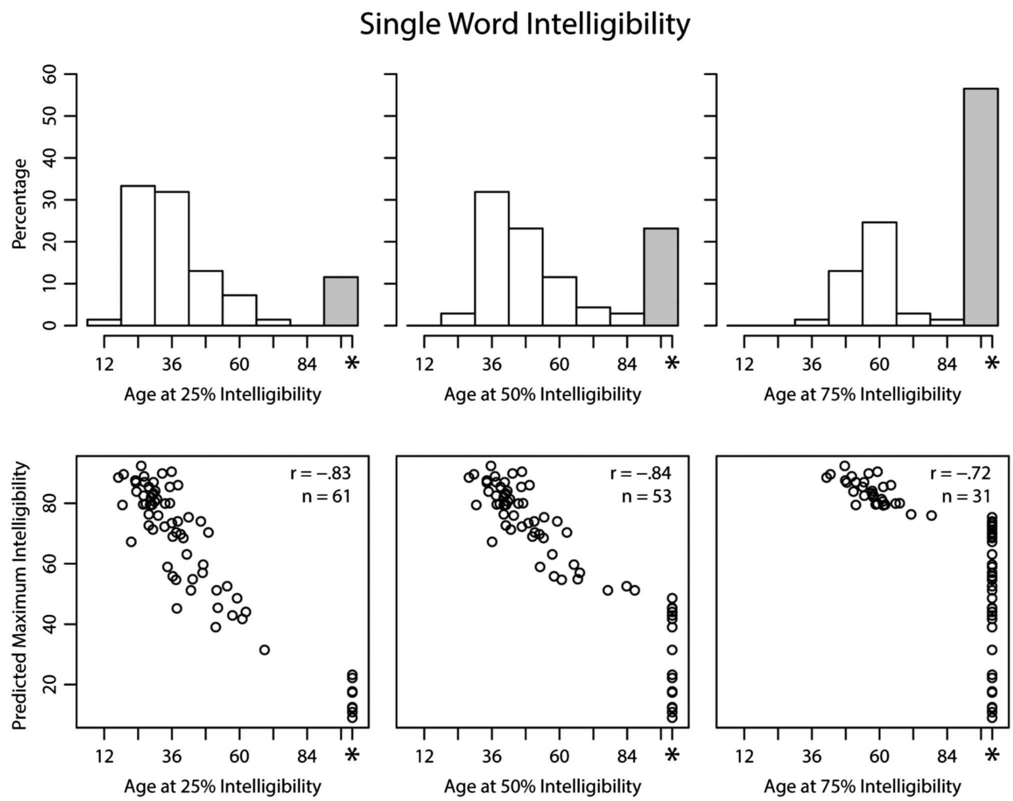
Second, children who cross intelligibility thresholds of 25%, 50%, and 75% at earlier ages have better outcomes when they are older; early performance is highly predictive of later outcomes.

Third, children with CP as a group have delayed speech intelligibility development, with only half of children reaching intelligibility levels of 73% or higher by the age of 96 months (8 years).

Finally, children with CP are still growing through 96 months (8 years), but growth is reduced after about 84 months (7 years). These findings are discussed in detail below.

1. The developmental window from 3 to 5 years of age seems to be an important time for intelligibility development in children with CP. During this time frame, intelligibility growth is the steepest—that is, children are growing/improving most quickly. Half of children have crossed the 25% intelligibility threshold by 36 months, and about half of children

Figure 3. Histograms and scatter plots of the distributions of age at crossing 25%, 50%, and 75% intelligibility thresholds. Note that histograms show percentage of children from the sample crossing target intelligibility thresholds at each age. Scatter plots show the correlations between the model-predicted maximum intelligibility and the age of crossing each intelligibly threshold. Note that (*) represents children who never crossed the target threshold in the 100-month range of the model.

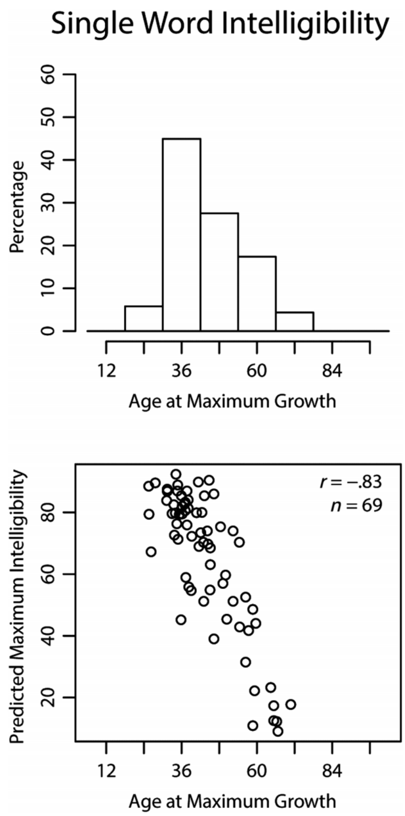


have crossed the 50% intelligibility threshold by 48 months. Few children with CP cross the 75% intelligibility threshold, but of those who do, most do so by about 60 months. Parallel data on typically developing children do not currently exist, so we cannot make direct comparisons regarding how children with CP compare as a group to typically developing peers in terms of intelligibility development. However, previous studies, some differing considerably methodologically, may provide a gross basis of comparison. Generally, the limited extant literature suggests that typical children may be between 53% and 75% intelligible at 3 years (Coplan & Gleason, 1988; Hodge & Gotzke, 2014a; Morris, Wilcox, & Schooling, 1995), 75% and 100% intelligible around 4 years (Coplan & Gleason, 1988; Hodge & Gotzke, 2014a; Morris et al., 1995; Wild, Vorperian, Kent, Bolt, & Austin, 2018), and between 82% and 90% intelligible around 5 years (Hodge & Gotzke, 2014a; Wild et al., 2018). Not surprisingly, findings for children with CP from this study suggest a considerable delay relative to age expectations, but it is possible that children with CP have similar rates of change to typical children. Data from typical children that are directly comparable are necessary to quantify whether the rate of change in intelligibility development in children with CP is consistent with the rate of change in intelligibility in typical children.

Several other lines of converging evidence related to early development in typical children generally suggest the period between 3 and 5 years is a time of considerable growth. For example anatomical growth of the vocal tract has been shown to be accelerated in the first 4–6 years of life, relative to later in development (Vorperian et al., 2009). Similarly, children between 4 and 5 years of age show considerable reduction in variability and increases in stability of speech motor control (Smith & Zelaznik, 2004). In addition, children are rapidly acquiring and nearing mastery of speech sounds, particularly consonants between 3 and 5 years of age. Specifically, nearly all consonant singletons are expected to have emerged by 5 years, and many have reached 90% mastery levels by 5 years (Sander, 1972; Smit, Hand, Freilinger, Bernthal, & Bird, 1990). Findings from this study are consistent with growth observed in other research.

Children in this study were receiving whatever speech and language treatment was provided to them in their usual environments throughout the time frame of this study. We gathered descriptive information regarding whether children were receiving speech-language treatment for the interval prior to each longitudinal visit. This information is summarized in Tables 2 and 3. Over the course of the study, about half of children were receiving therapy at any given time point.

Figure 4. Histogram and scatter plot of the distribution of age of maximum growth in speech intelligibility. Note that the histogram shows the percentage of children from the sample reaching the steepest growth by age. The scatter plot shows the correlation between the model-predicted maximum intelligibility and age at maximum growth. All children are included in the results.



This was true for each time point between 3 and 5 years of age. Examination of the effects of treatment on change in speech intelligibility is well beyond the scope of this article. We can only conclude that intelligibility changes occurred, perhaps because children were developmentally primed for growth. Any treatment that occurred may have been enhanced by this developmental propensity for growth, but we cannot separate with our current data set the relative contributions of intervention to growth.

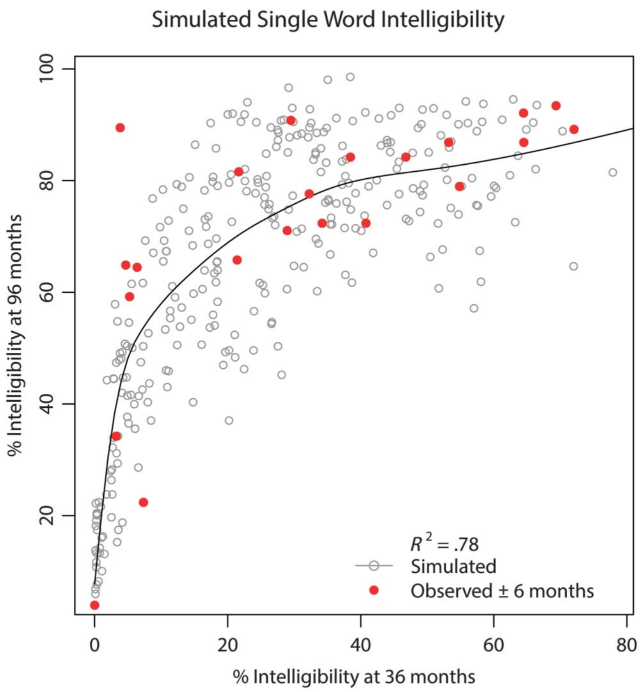
In this study, we did not differentiate among children with CP who had speech motor impairment and those who did not have speech motor impairment. One challenge with early speech development in children with CP is making this differential diagnosis, particularly for children who may have more subtle issues, for example, those who have mild or even questionable speech motor involvement. A key problem is that features of early typical speech development overlap with features of speech motor impairment (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 speech motor impairment in children with CP (Hustad et al., 2010, 2016). Among children examined in this study, Table 1 shows that 23 children did not have speech motor impairment at 4 years of age, and the other 46 children did have speech motor impairment. Because of the stable nature of the neurological involvement that underlies CP, children do not tend to outgrow speech motor impairment, although speech characteristics do change with development. One important question is whether trajectories of development and developmental outcomes differed for children with speech motor impairment and children without speech motor impairment. Although we did not formally examine this question in this article, data descriptively suggest that children without speech motor impairment had the highest intelligibility scores at 96 months of age. However, longitudinal data plotted in Figures 1 and 2 suggest a continuum of individual differences in growth, including children who made consistent and rapid change over time and children who never surpassed 20% intelligibility. Future work will seek to examine empirically differences in growth among children and whether the age of peak growth is impacted by the presence of speech motor impairment.

2. Children who cross intelligibility thresholds earlier in life have better outcomes later; early performance is highly predictive of later outcomes. In this study, two findings support this conclusion. First, maximum achieved intelligibility was highly negatively correlated with the age of crossing each intelligibility threshold (25%, 50%, and 75%). Specifically, the younger children were when they crossed each intelligibility threshold, the higher the maximum achieved intelligibility at 96 months. Second, when we examined simulation results based on model parameters from observed intelligibility development data, we found that intelligibility at 3 years was highly predictive of later intelligibility scores at 96 months; specifically, 78% of the variance in simulated intelligibility scores at 96 months was explained by performance at 36 months. Descriptively, this was especially true for children in the 10%–40% intelligibility range at 3 years, highlighting particular relevance to the children who are more likely to have speech motor impairment (as indicated by lower speech intelligibility scores). Both findings are consistent with our earlier work demonstrating that early performance is highly predictive of later outcomes in children with CP (Hustad et al., 2017, 2018). Findings are also consistent with results from other domains such as gross motor development, which indicate that early abilities predict later outcomes in children with CP (Kolobe, Bulanda, & Susman, 2004; Rosenbaum et al., 2002).

From a clinical perspective, this finding has potential implications for treatment. It is possible that early therapy focused on speech production and intelligibility, capitalizing on a period of rapid developmental change and getting children to intelligibility benchmarks at earlier ages may change developmental trajectories and long-term outcomes. Research is necessary to determine if this is true, but the potential is promising.

3. Children with CP overall as a group have delayed speech intelligibility development. Although definitive

Figure 5. Scatter plot showing prediction of 96-month intelligibility outcomes from 36-month single word intelligibility data. Data were obtained using single word intelligibility data simulations based on models depicted in Figure 2. Note that red dots represent data from observed children and open dots represent data from simulations.



normative data for intelligibility development in typical children have not been established for single word intelligibility as measured by naïve listeners, findings from the limited extant literature suggest that typical children, on average, might be expected to have about 90% single word intelligibility between 5 and 7 years of age (Wild et al., 2018), whereas 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 this study, only 25% of the children with CP reached intelligibility expectations for typical 4- to 5-year-olds by 8 years of age. Specifically, 82% intelligibility corresponded to the 75th percentile mark at 96 months of age. Our results further showed that none of the children reached 100% intelligibility by 96 months of age (although one child came close), even though a third of the children did not have clinical dysarthria.

The 50th percentile mark for maximum intelligibility at 96 months was 73%, indicating that half of children were above this point and half were below. The 25th percentile mark for maximum intelligibility was 53%, indicating that a majority of children reached a 50% intelligibility threshold. It is important to note, however, that intelligibility of 50% or even 70% at 8 years of age represents a considerable deficit that could be characterized as moderately–severely impaired from a clinical speech motor disorders perspective. It is very likely that the large proportion of children in this intelligibility range would have significant difficulty in communicating using speech alone and would therefore benefit from multimodal communication options including AAC systems and strategies to support communication.

Further research is necessary to understand how children with speech motor involvement differ from those without speech motor involvement in their outcomes. In particular, the question of whether and when children without speech motor impairment diverge in their intelligibility development from those with speech motor impairment is an important one that has intervention implications and therefore should be addressed.

4. Children with CP are still growing through 96 months (8 years), but growth is reduced after about 84 months (7 years). In this study, we followed children longitudinally until 96 months of age and our predictive models extrapolated the data to 100 months of age. Most children continued to show changes in intelligibility throughout the time course of this study; however, growth slowed dramatically after 7 years of age. Research on speech motor development suggests that motor control continues to improve, becoming more stable and less variable through adolescence (Smith & Zelaznik, 2004). Furthermore, recent work in Down syndrome has revealed that intelligibility continues improving through 16 years of age (Wild et al., 2018). Whether children with CP experience another growth spurt in speech development after 96 months of age and the extent to which they continue to make slow change beyond this time are unknown because longitudinal work examining intelligibility of children with CP into adolescence has not been conducted previously. Such work is necessary to complete our understanding of the limits of change in speech development and to identify other periods of rapid change when treatment may be particularly beneficial in changing growth trajectories and outcomes. Results of this study suggest that there continues to be room for improvement for most children with CP, with only half of children reaching 73% intelligibility by 96 months of age. These results also highlight the importance of AAC interventions to support or supplement speech for the 50% of children with CP who are able to speak but do not surpass 75% intelligibility thresholds during the school-age years.

Limitations and Future Directions

There were a number of limitations to this study. Children with CP were not separated according to whether or not they had speech motor involvement (dysarthria) in this study. Rather, all children were considered together to describe the distribution of growth patterns across the population. Further analyses are necessary to examine trends and growth trajectories by speech motor status. One important observation, however, is that descriptive data plotted in Figures 1 and 2 suggest a continuum of growth trajectories across children.

Perhaps the most significant limitation of the present research is that parallel data on typically developing children have not been published. As a result, it is difficult to interpret our findings relative to typical age expectations

Table 3. Number of intelligibility observations by 6-month age band.

Age band in months	No. of visits	Age in months, *M* (*SD*)	Intelligibility *M* (*SD*)	Intelligibility Range	Visits with concurrent therapy
24–29	8	27 (2)	24% (14)	9%–47%	4
30–35	24	33 (2)	25% (17)	0%–64%	18
36–41	37	39 (2)	36% (20)	3%–72%	21
42–47	59	44 (2)	42% (23)	0%–82%	27
48–53	61	50 (2)	53% (22)	1%–89%	30
54–59	67	56 (2)	56% (25)	0%–91%	36
60–65	65	63 (2)	58% (25)	2%–91%	34
66–71	59	69 (2)	58% (27)	0%–93%	26
72–77	57	74 (2)	63% (27)	3%–95%	26
78–83	61	80 (2)	62% (26)	4%–95%	21
84–89	35	86 (2)	69% (22)	8%–93%	19
90–96	33	94 (2)	63% (28)	1%–99%	18
Total	566				280

and to quantify the extent to which growth trajectories are similar to and different from typical children. Analogous data on typical children would allow us to begin to develop benchmarks and cut-points for typical versus atypical intelligibility development in children with CP and other populations of children with speech disorders.

Intelligibility is a complex and multifaceted construct that is critically important in the field of speech-language pathology. In this study, we examined only single word intelligibility. We did not examine phonetic or acoustic features of the speech of children with CP. This type of data could provide important information about production features of speech and could inform the underlying bases of intelligibility deficits (Wild et al., 2018), perhaps pointing to potential intervention targets. Analyses of this nature are currently underway and will be used to further refine our understanding of the ways that CP impacts production features of speech and how these features change over time in children.

As discussed in the introduction section, single words are generally more difficult for listeners to understand than connected speech. In addition, we used a highly controlled experimental paradigm involving unfamiliar listeners to measure intelligibility in a quiet laboratory environment. Listeners did not have the benefit of contextual information that would be present in a real interaction, they did not have the opportunity to see the children as listeners in a real environment would, and they did not have to contend with background noise that is present in most interactions. Thus, results presented here may not provide a comprehensive representation of intelligibility. Studies should examine intelligibility of connected speech and the extent to which single word and multiword findings are similar and different. Until such information is available, we suggest that the single word intelligibility data presented in this article reflect only part of the picture of intelligibility development.

Clinical Implications

In spite of its limitations, there are a number of potential clinical implications from this study. First, on the later end of the age range for this study, nearly all children with CP who were able to speak had reduced intelligibility at 8 years of age, and very few children exceeded 83% intelligibility; in fact, more than half of the children in this study did not reach 75% intelligibility for single words. On the early end of the age range for this study, children who reached 25%, 50%, and 75% intelligibility thresholds at earlier ages had better long-term intelligibility outcomes, as evidenced by extremely strong correlations. The time between 3 and 5 years of age was a window of considerable growth in speech intelligibility, when children were changing the most rapidly. About half of the children in this study were receiving speech-language intervention at any given time point in this study. However, we do not know the specific nature or frequency of intervention, nor do we have data regarding children's progress in treatment. Therefore, we do not know if the interventions that children were receiving may have magnified the changes we observed in speech intelligibility in this study. Determining whether treatment during periods of rapid growth may accelerate that growth further is an important area for further investigation. Intervention in this window may serve to accelerate changes even further and result in better outcomes later. One potential set of guidelines that are broadly inclusive, focusing on 25th percentile findings for 25%, 50%, and 75% intelligibility thresholds are as follows: (a) Children who have not reached 25% single word intelligibility by 29 months of age should be considered for speech therapy if they are not already receiving it to enhance early intelligibility development and to introduce multimodal communication to foster expressive communication. Beginning treatment by 29 months would ensure that children are receiving therapy when they enter a period of rapid speech development at 3 years. (b) Similarly, for children who are not already receiving speech therapy and who have not reached 50% intelligibility by 40 months of age, implementation of speech therapy that focuses primarily on multimodal communication and includes AAC systems and strategies to support and/or supplement speech should be considered. (c) Children who have not reached 75% intelligibility by 58 months of age should be considered for AAC focused intervention to ensure that they have access to expressive modes of communication to support educational participation. Intelligibility focused therapy may still be beneficial, but as children enter a reduction in rate of growth after 5 years, progress may be slower with regard to change in speech. Note, however, that communication needs will become more complex and multifaceted as language and cognitive abilities advance with development; thus, access to tools that enable full participation is critical and should be a priority in therapy.

These clinical implications are based on our interpretation of growth results from a large set of prospective longitudinal quantitative evidence. Data of this nature have never been obtained previously, to our knowledge. We apply our clinical experience/expertise and firsthand knowledge over years of study of the children included in this research, which comprise an important part of evidence-based practice

(Dollaghan, 2007; Ratner, 2006) to the clinical implications presented here.

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