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Research Article

Longitudinal Growth in Intelligibility of Connected Speech From 2 to 8 Years in Children With Cerebral Palsy: A Novel Bayesian Approach

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Aim: The aim of the study was to examine longitudinal growth in intelligibility in connected speech from 2 to 8 years of age in children with cerebral palsy.
Method: Sixty-five children with cerebral palsy participated in the longitudinal study. Children were classified into speech-language profile groups using age-4 data: no speech motor impairment (SMI), SMI with typical language comprehension, and SMI with impaired language comprehension. We fit a Bayesian nonlinear mixed-effects model of intelligibility growth at the child and group levels. We compared groups by age of steepest growth, maximum growth rate, and predicted intelligibility at 8 years of age.
Results: The no SMI group showed earlier and steeper intelligibility growth and higher average outcomes compared to the SMI groups. The SMI groups had more variable growth trajectories, but the SMI with typical language comprehension group had higher age-8 outcomes and steeper rates of maximum growth than the SMI with impaired language comprehension group. Language comprehension impairment at age of 4 years predicted lower intelligibility outcomes at age of 8 years, compared to typical language at age of 4 years.
Interpretation: Children with SMI at age of 4 years show highly variable intelligibility growth trajectories, and comorbid language comprehension impairment predicts lower intelligibility outcomes.
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Speech intelligibility is a critical concern for children with cerebral palsy (CP). Recent studies suggest that approximately 50% of children with CP have the speech motor impairment (SMI) dysarthria (Nordberg et al., 2013). Dysarthria is a manifestation of central or peripheral neuropathology affecting structures involved in motor control of the speech subsystems (articulation, phonation, respiration, and resonance; Darley et al., 1969; Duffy, 2005). Reductions in speech intelligibility are ubiquitous in dysarthria and can have a critical impact on educational, social, and vocational participation (Dickinson et al., 2007; Fauconnier et al., 2009).

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Intelligibility is 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 dyadic, with both listener and speaker making joint contributions. Intelligibility can be influenced by many variables, including the length and nature of speech materials being produced (e.g., single words vs. multiword sentences; Kent et al., 1994). Important clinical decisions are often made based on speech intelligibility measures from children. For example, intelligibility scores can serve as a basis of comparison for documenting and monitoring change in speech performance (Yorkston et al., 1999), as an index of severity of the dysarthria (Weismer & Martin, 1992), and as an indicator of functional ability (or disability) relative to "normal" performance (Yorkston et al., 1999). Intelligibility scores may also guide treatment decision making about provision of augmentative and alternative communication tools and technologies to support functional communication when speech is not sufficient to meet communication needs across partners and contexts.

Little has been known about early predictors of later speech development in children with CP until recently.

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However, longitudinal studies reveal that variables such as age of onset of word production (Hustad et al., 2019) and single-word intelligibility at 3 years of age are both highly predictive of later speech outcomes in children with CP (Hustad et al., 2018). This work also suggests that, when children with CP are separated into speech-language profile groups (SLPGs), as reported by Hustad, Mahr, Broman, and Rathouz (2020), profile-specific differences are observed for age of crossing intelligibility quartile thresholds, age of most rapid growth, and intelligibility thresholds at 8 years of age. In addition, prediction of single-word intelligibility outcomes at 8 years of age based on data from children at 3 years of age is improved when speech-language profile status is included as a predictor. In this work on SLPGs, a key differentiator among groups is the presence or absence of SMI and the presence or absence of language comprehension impairment. We have used language comprehension as a general proxy for cognitive abilities (Hustad et al., 2010), and recent work suggests very high convergence between IQ scores and language comprehension scores in children with CP (Soriano & Hustad, 2020). Furthermore, research suggests that there are complex interactions between language development, cognitive development, and speech development (Nip & Green, 2013). In particular, children with language impairment may experience differential negative penalties in their speech motor control relative to those without concomitant language impairment (Vuolo & Goffman, 2018). In this study, we were interested in examining how intelligibility development for connected speech might be impacted by the presence of language/cognitive involvement in children with CP.

Previous research has provided clear evidence that single-word intelligibility is different than intelligibility of connected speech in adults. Generally, single words tend to be less intelligible because there is less linguistic context to aid listeners (Miller et al., 1951). However, for typically developing children, recent work suggests that single-word intelligibility tends to be higher than multiword intelligibility until about 40 months of age, at which point multiword intelligibility becomes higher than single-word intelligibility (Hustad, Mahr, Natzke, & Rathouz, 2020). The impact of growth over time on multiword and single-word intelligibility is unknown for children with dysarthria, particularly those with moderate or severe intelligibility deficits. One possibility is that the increased speech motor control demands of multiword utterances may have a persistent detrimental impact on intelligibility (Hustad et al., 2012). Alternatively, the contextual information provided in multiword utterances may lead to an advantage for children with dysarthria because listeners are able to access signal-independent knowledge as they try to make sense of the speech signal. Regardless, multiword utterances have high ecological validity and represent how children communicate in everyday life. Intelligibility scores from multiword productions provide a useful indicator of functional speech production abilities. Thus, it is critical to examine longitudinal intelligibility growth in multiword utterances among children with CP over a protracted time.

This study reports the longitudinal development of intelligibility based on productions of multiword utterances for a cohort of children with CP. We split the children into three SLPGs based on their speech and language status at age of 4 years, following our previous work (Hustad, Mahr, Broman, & Rathouz, 2020): no SMI (NSMI), SMI with typical language comprehension (SMI-LCT), and SMI with impaired language comprehension (SMI-LCI). We expected children in all three groups to be heterogeneous, but we also expected that profile groups would capture similarities among the children within groups and that there would be reliable group differences. We also expected speech and language status to predict age-8 outcomes, so that our three groups would follow the ordering NSMI > SMI-LCT > SMI-LCI. That is, children without SMI at age of 4 years would have higher expected intelligibility outcomes than children with SMI, and children with SMI and impaired language comprehension would have lower outcomes on average than children with SMI and typical comprehension. We addressed the following specific research questions.

1. Do children with different speech-language profiles show different patterns of intelligibility growth in their connected speech? In particular, we examined
   a. age of steepest growth,
   b. rate of steepest growth, and
   c. intelligibility at 8 years.

2. How does the presence of language comprehension impairment influence predicted intelligibility growth trajectories for children with SMI?

Question 1 asks a conventional data analysis question: How do these groups of participants differ for these measures (intelligibility growth features)? Question 2 asks a different sort of question about clinical forecasting: Given what we learned for Question 1, what intelligibility growth trajectories are plausible based on a single intelligibility measurement at age of 4 years?

Method

Participants

Children with CP were selected from a larger longitudinal study of communication development in children with CP; thus, they are a subset of those described in Hustad, Mahr, Broman, and Rathouz (2020). Inclusion in the longitudinal study required a medical diagnosis of CP and normal hearing as confirmed by audiological evaluation or screening with distortion product otoacoustic emissions. From this larger cohort, we selected children who contributed at least two visits to the longitudinal study, could produce utterances in our connected speech task of at least two words in length, and did not have a concomitant autism spectrum diagnosis. The study obtained ethical approval from the University of Wisconsin–Madison Institutional Review Board for Social and Behavioral Sciences. All parents provided informed consent for participation in the study.

A total of 65 children (31 girls, 34 boys) met our criteria and were included in the data set presented here. Note that three of the children from the earlier articles were unable to produce utterances two words in length or longer and thus were excluded from the present analysis. Each child contributed 2–12 data points, for a total of 513 data points across the 65 children, yielding a mean of 7.9 (*SD* = 2.3) and a median of 8 data points per participant. Data points were obtained at 6-month intervals to 8 years of age. However, children began the study at different ages, and even after beginning the study, not all children were able to contribute data points every 6 months due to health issues, family scheduling challenges, and attrition. See Table 2 for a summary of the number of visits per child per profile group. 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. Table 2 provides a summary of how many children were enrolled in speech-language therapy by profile group during each visit represented in this article.

Adults Listeners

A total of 1,026 normal-hearing adults served as listeners in this study. Two different listeners heard the speech of each child at each visit (513 child visits × 2 listeners per child = 1,026 listeners); each listener heard only one child producing all stimulus material. Listeners were recruited from the university community via public postings and were primarily undergraduate students. All listeners reported no more than incidental experience listening to or communicating with persons having communication disorders, and all reported a negative history of language, learning, or cognitive disability. Listeners were compensated monetarily for their participation. They were 268 male and 758 female listeners. The mean age of listeners was 20.8 years (*SD* = 5.7).

Note that children and their adult listeners are the same as those described in Hustad et al. (2019) and Hustad, Mahr, Broman, and Rathouz (2020) for single-word intelligibility. Data presented in the current article reflect multiword intelligibility results, which were not included in the earlier articles.

Materials and Procedure

For the larger longitudinal study, a speech-language evaluation protocol was administered by a research speech-language pathologist (SLP) in a sound-attenuating suite. For this study, multiword intelligibility was of interest. We also collected language comprehension data, which we used to classify children into SLPGs, described below.

Speech Intelligibility

We obtained multiword intelligibility measures by having children produce a corpus of speech stimuli, which were the same for each child and each longitudinal visit.

Table 1. Demographic characteristics of participants.

Characteristic	NSMI (*n* = 22)	SMI-LCT (*n* = 31)	SMI-LCI (*n* = 12)
Male/female ratio	17:5	11:20	6:6
No. of visits, *M* (*SD*)	8.7 (2.0)	8.0 (2.2)	6.2 (2.2)
No. of visits, *Mdn*	8.5	8.0	5.5
Age at first visit (months), *M* (*SD*)	41 (8)	43 (10)	53 (15)
Age at last visit (months), *M* (*SD*)	88 (10)	88 (10)	86 (9)
GMFCS (Palisano et al., 1997)			
I	19	14	5
II	3	4	1
III	0	5	1
IV	0	7	3
V	0	1	0
(Missing)	0	0	2
CP type			
Spastic			
Diplegia	6	6	1
Hemiplegia (left)	9	4	1
Hemiplegia (right)	4	8	3
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	1	1	3

Note. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment with typical language comprehension; SMI-LCI = speech motor impairment with impaired language comprehension; GMFCS = gross motor function classification system; CP = cerebral palsy.

Table 2. Visits and therapy services by age by profile group.

Age band in months	Profile group	Visits	Visits with concurrent therapy	Age in months, *M* (*SD*)	Intelligibility, *M* (*SD*)	Intelligibility, range
24–29	NSMI	1	0	25	44%	—
	SMI-LCT	1	1	24	17%	—
	SMI-LCI	0	0	—	—	—
30–35	NSMI	6	3	33 (2)	36% (17)	16%–65%
	SMI-LCT	8	5	33 (2)	18% (15)	0%–41%
	SMI-LCI	0	0	—	—	—
36–41	NSMI	11	6	38 (2)	46% (16)	21%–67%
	SMI-LCT	13	6	39 (2)	20% (18)	0%–59%
	SMI-LCI	4	4	39 (1)	20% (14)	3%–32%
42–47	NSMI	18	6	44 (2)	60% (17)	28%–84%
	SMI-LCT	22	10	44 (2)	25% (16)	0%–55%
	SMI-LCI	6	5	45 (2)	26% (15)	10%–46%
48–53	NSMI	23	4	51 (2)	69% (14)	43%–89%
	SMI-LCT	26	18	50 (2)	34% (19)	2%–72%
	SMI-LCI	6	6	50 (2)	28% (21)	5%–55%
54–59	NSMI	25	6	57 (2)	80% (9)	56%–95%
	SMI-LCT	30	22	56 (2)	41% (24)	0%–84%
	SMI-LCI	8	8	56 (2)	23% (18)	4%–60%
60–65	NSMI	22	4	63 (2)	85% (9)	63%–95%
	SMI-LCT	30	20	62 (2)	45% (22)	0%–87%
	SMI-LCI	11	11	63 (2)	33% (19)	0%–69%
66–71	NSMI	19	1	69 (2)	86% (8)	69%–98%
	SMI-LCT	27	14	68 (2)	54% (28)	0%–89%
	SMI-LCI	10	7	69 (2)	28% (22)	3%–75%
72–77	NSMI	20	1	74 (2)	90% (6)	76%–98%
	SMI-LCT	27	14	74 (2)	61% (29)	1%–94%
	SMI-LCI	9	7	74 (2)	25% (16)	0%–45%
78–83	NSMI	22	2	81 (2)	91% (7)	66%–98%
	SMI-LCT	29	17	80 (2)	56% (29)	0%–93%
	SMI-LCI	10	7	80 (2)	22% (16)	1%–42%
84–89	NSMI	13	1	87 (2)	92% (5)	83%–99%
	SMI-LCT	18	9	86 (2)	72% (23)	2%–98%
	SMI-LCI	5	3	87 (1)	21% (21)	0%–51%
90–96	NSMI	11	0	94 (2)	96% (2)	93%–99%
	SMI-LCT	17	11	94 (2)	61% (36)	1%–96%
	SMI-LCI	5	4	95 (2)	28% (18)	4%–44%
Total	NSMI	191	34			
	SMI-LCT	247	147			
	SMI-LCI	74	62			

Note. Em dashes indicate that the statistic cannot be computed because the the number of data points is equal to 0 or 1. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment with typical language comprehension; SMI-LCI = speech motor impairment with impaired language comprehension.

We used an iPad to present each child with an image and a prerecorded auditory model, which was immediately repeated by the child. Multiword utterances were taken from the Test of Children's Speech (TOCS+; Hodge & Daniels, 2007) and comprised 60 sentences ranging from two to seven words (10 of each sentence length). Lexical, phonetic, syntactic, and morphological features of all stimuli were developed to be appropriate for children. The multiword protocol started with the 10 two-word utterances and advanced to three-word utterances and so on—stopping when the child was not able to go any further. Speech samples from children were recorded using a digital audio recorder (Marantz PMD 570) 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.

Digital recordings of children's speech were edited to remove any extraneous noises and were peak amplitude-normalized for playback to listeners. Speech samples were presented to listeners individually in a sound-attenuating field with peak audio output levels calibrated to approximately 75 dB SPL. Utterances produced by children were randomized for each listener and were played only once. Listeners were instructed that children would be producing real words and instructed to take their best guess if they were unsure what the child said. Listeners typed what they thought the child said into an in-house software application.

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 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 et al., 2019). We used an average-score, consistency-based, one-way random effects model, and we found strong agreement for the 513 listener pairs, ICC(2) = .989, 95% CI [.987, .991]. Moreover, the average difference between the two listeners of each child for each visit was 4.5 percentage points (*SD* = 4.1).

Language Comprehension

The following measures were administered, depending on the child's age, developmental level, and motor skill profile: Test for Auditory Comprehension of Language–Third Edition (Carrow-Woolfolk, 1999), Preschool Language Scales–Fourth Edition (Zimmerman et al., 2002), and Peabody Picture Vocabulary Test–Fourth Edition (Dunn & Dunn, 2007). Standard administration procedures were adapted to enable participation in testing for items involving manual manipulation. We obtained standard scores for each administration of a test. Children who had language scores more than 1.5 *SD*s below age expectations were classified as having language comprehension impairment per the manual of respective tests (Carrow-Woolfolk, 1999; Dunn & Dunn, 2007; Zimmerman et al., 2002).

Classification Into Profile Groups

At 48–54 months, children were classified into one of three SLPGs following our earlier work (Hustad et al., 2010, 2016). We selected this age range because we have been able to reliably diagnose SMI in children with CP at this age (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. Diagnosis of SMI prior to 48 months is challenging and often impossible because children with CP tend to be delayed in 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).

Classification methods have been described in detail previously (Hustad, Mahr, Broman, & Rathouz, 2020). Briefly, each child was independently classified by two SLPs based on clinical judgment of the presence or absence of SMI and the presence or absence of language comprehension impairment. Children who were classified as having no speech motor involvement (NSMI) had no clinical evidence of speech impairment based on clinician observation during the data collection session and were confirmed via review of video and audio recordings after the session. Only one child within the NSMI group had language comprehension scores that were outside the range of typical expectations. Children who were classified as having speech motor involvement (SMI), by definition, had clinical evidence of dysarthria, which was determined through clinical observation of the presence or absence of dysarthria features, 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 during administration of the TOCS+ and during a spontaneous speech sample. Binary judgments of typical language comprehension and impaired language comprehension were made based on results of standardized testing, described above. We focused on only language comprehension and not on expressive language because it was more amenable to measurement and modifications could be made to accommodate motor impairment in the administration of language comprehension testing. Expressive language measurement is more challenging because of the high prevalence of SMI and associated intelligibility deficits that compromise the reliability and validity of scoring when definitive spoken targets are not known (as in the case of language sample analysis). Children who had language comprehension impairment and SMI were classified as SMI-LCI; those who had language comprehension that was typically developing and had SMI were classified as SMI-LCT. Classification agreement between the two SLP raters was 100%.

Statistical Analyses

Bayesian Preliminaries

We used Bayesian regression models to estimate intelligibility growth trajectories for each child, along with the average growth trajectory across children at the profile-group level, and to *quantify the uncertainty* about the estimated growth trajectories. The goal was to obtain a range of plausible values for growth parameters given the data. For example, suppose we wanted to learn about the age when growth was steepest on average for children in the NSMI group, and we would also like to get a sense of how uncertain we are about this estimated age. The Bayesian models address this need by letting us ask and answer questions such as: What is the probability, given the data, that this true age falls before 48 months on average? What is this probability for a specific child, given the data? What range of ages that has a 95% probability, given the data, of containing the true age of maximum growth?

To frame the Bayesian approach, note that *all statistical modeling* approaches start by positing a general data-generating process—called a model—that is governed by statistical parameters. These parameters (a) are unknown; (b) usually capture the scientific, policy, or clinical question we are trying to answer with the data; and (c) are the targets of statistical inference. A common example of a parameter is the slope in a regression model for the mean test score as a function of age. A model will also contain other parameters to complete the picture (e.g., in linear regression, we also estimate a constant term and the standard deviation of the error term). The Bayesian approach then deviates from the more common frequentist approach in that it also requires specification of uncertainty about each parameter before the data are analyzed; this is done in terms of a *prior distribution* (i.e., *before* the data).

The modeling work then combines the prior with the data to obtain a *posterior distribution* for the model parameters we care about. The posterior distribution captures the distribution of plausible parameter values in the model given the data. As such, statistical inferences about those parameters are framed in terms of probability statements describing the posterior distribution. A standard single-number summary of the distribution (a point estimate) is the median. We can estimate the probability that a parameter falls in a given range or the probability the direction of an effect is positive or negative. We can also obtain a range of values for the parameter that captures 95% of the posterior probability. This last object is called a "posterior (credible) interval" and is loosely used similarly to a confidence interval in classical frequentist inference. (These two intervals have different meanings, but in many situations, the two will largely coincide.) Kruschke and Liddell (2018) provide a review contrasting Bayesian versus frequentist approaches. Lambert (2018) and McElreath (2020) provide textbook treatments on Bayesian statistics.

Current Analyses

For Research Question 1, our analysis goal was to model growth in intelligibility at the group level and child level. That is, we sought to estimate and compare both the *average* growth trajectories in each group and the *population variation* among growth trajectories across children within each group. This population variation quantifies the range of typical variation in trajectories within each group, which also corresponds to the predicted developmental trajectories for a new, as-yet unobserved child.

To perform our analysis, we used a Bayesian nonlinear mixed-effects beta regression model to estimate how the percentage of intelligible words changes with age. The full parameterization of the model is provided in Supplemental Material S1. The key features of the model are as follows: a nonlinear template curve for growth trajectories, the estimation of average curve parameters in each group, and the estimation of between-children (population) variation in curve parameters in each group. More specifically, we modeled growth with a logistic curve that served as the template for intelligibility growth. This logistic curve used three parameters: an *asymptote,* or ceiling, for intelligibility growth at the higher ages (which we expect to be somewhat near 100% for some but not nearly all children); a *midpoint* age when growth is steepest; and a *scale* factor, which sets the curve's slope. For each SLPG, we estimated the average of these growth curve parameters and hence the average developmental trajectory for each group. We also gave participants their own asymptote, midpoint, and scale values by estimating by-child random components for these three parameters. We estimated random-effect variances and covariances for each group; this setup allowed the amount of between-children variability in the asymptotes, midpoints, and scales to change from group to group. The beta regression model also included a precision parameter that estimated within-child variability (or degree of measurement noise) over time. We allowed this precision parameter to change linearly with age and by profile group.

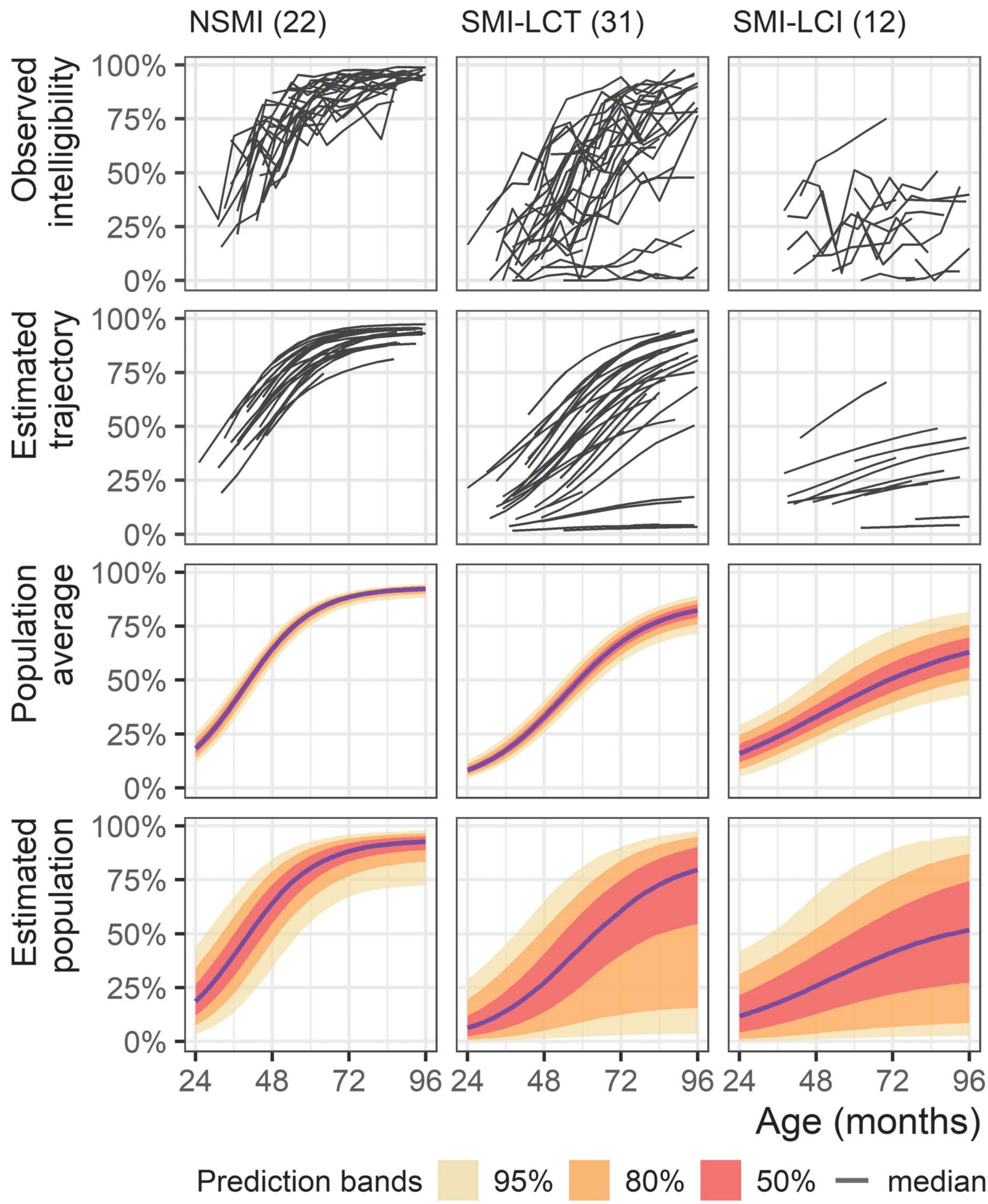
For Research Question 2, we took advantage of the information borrowing of mixed models to perform a novel kind of clinical forecasting. Our Bayesian mixed-effects model worked on two levels: It characterized the population of growth trajectories within each group, and for each child, it also estimated the distribution of plausible growth trajectories. For a child with many observations, the growth trajectories would closely follow the data, but for a child with relatively few observations, the path of estimated growth trajectories before or after the observed intelligibility scores would "borrow information" from the group-level population. This feature of the model meant that we could estimate a distribution for growth trajectories from *a single intelligibility observation,* and these trajectories would be consistent with that observation and the typical variation in growth trajectories in each profile group. That was our approach for Question 2: We considered a hypothetical new child who was seen clinically at age of 4 years, placed in one of the three profile groups, and assessed for intelligibility at age of 4 years as well. Based on our fitted model, we estimated what growth trajectories and intelligibility scores at age of 8 years would be plausible given those two pieces of information. This exercise was used to examine and illustrate how the presence of language comprehension impairment influences intelligibility outcomes.

Our analysis was carried out in the R programming language (Version 3.6.3; R Core Team, 2020) using the brms interface (Version 2.12.0; Bürkner, 2017) to the Stan programming language (Version 2.19.3; Carpenter et al., 2017). Posterior summary, prediction, and visualization were facilitated using the tidybayes package (Version 2.0.3; Kay, 2019). We sampled the posterior distribution using Hamiltonian Monte Carlo with four chains and 2,000 post-warm-up samples per chain. The model passed all diagnostic statistics (all $\hat{R}$ values < 1.01, all effective sample sizes > 400, zero divergent iterations).

Results

Figure 1 shows observed intelligibility scores (top row), each child's posterior median growth curve (second row), estimated population-average growth curves from each group (third row), and estimated population variation—specifically, the intelligibility predictions for a new, as-yet unobserved child in each group (fourth row). One can think of the population average and its posterior prediction band (third row) like the uncertainty quantified by a mean and confidence intervals: Adding more participants to each group would reduce the standard error and provide tighter posterior prediction bands. In contrast, the

Figure 1. Observed intelligibility scores and estimated growth trajectories. First row: Observed intelligibility with one connected line per child. Second row: Estimated trajectories for each child based on Bayesian model fit. Our model estimated a distribution of lines for each child, but for ease of presentation, we show lines computed using the posterior medians of each child's growth curve parameters. Third row: Estimated population average of growth trajectories. Prediction bands here quantify uncertainty about the average trajectory (similar to 95%, 80%, and 50% confidence intervals). Groups with fewer children or more variability have wider prediction bands. Fourth row: Estimated population variability of individual growth trajectories. Prediction bands here quantify the population variability in trajectories about the average (similar to a standard deviation). Specifically, these bands show the predicted trajectories for a new, as-yet unobserved child in each group. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment with typical language comprehension; SMI-LCI = speech motor impairment with impaired language comprehension.



population predictions (fourth row) are analogous to the uncertainty quantified by a mean and standard deviation. Adding more participants would improve the precision with which the mean and variance are estimated, but it would not change the spread of typical values observed in each group. Table 2 reports summary statistics on the observed intelligibility scores by group in 6-month age increments. Table 3 provides numerical summaries of key model estimates including group means and group differences.

Question 1: Do Children With Different Speech-Language Profiles Show Different Patterns of Intelligibility Growth in Their Connected Speech?

Age of Steepest Growth

The logistic growth curve model assumes that children start at a floor of 0% intelligibility and grow toward some asymptotic level. From the floor to the midpoint, growth is accelerating, and from the midpoint to the ceiling, growth decelerates with diminishing gains with age. This midpoint marks an inflection point where growth is steepest.

The estimated average ages of steepest growth for each group were as follows: NSMI, 39 months [95% posterior interval: 36, 42]; SMI-LCT, 54 months [50, 59]; SMI-LCI, 51 months [33, 72]. Children in the NSMI group had their age of steepest growth at least a year ahead of their peers in the SMI-LCT group: $\text{mid}_{\text{NSMI}} - \text{mid}_{\text{SMI-LCT}} = -15$ [−21, −10]. The SMI-LCI group's smaller sample size and the variability among the children in the group made the estimated group average highly uncertain. The 95% posterior intervals for the NSMI and SMI-LCT groups spanned 6 and 9 months, respectively; the interval for SMI-LCI group spanned 38 months. This uncertainty in the group average propagated to the comparisons between groups, so the differences between the SMI-LCI group and others were also unclear.

There was likely less between-children variability in the growth curve midpoints for the NSMI group compared to the two SMI groups. For the NSMI group, the standard deviation for the population variation in the midpoint was 6.7 months [4.3, 10.0]. In the SMI groups, standard deviations of 10 months or larger were plausible, $SD(\text{mid})_{\text{SMI-LCT}} = 10.6$ [7.7, 13.9], $SD(\text{mid})_{\text{SMI-LCI}} = 11.0$ [5.9, 16.0].

Rate of Steepest Growth

The estimated average growth rates at the age of steepest growth for each group were as follows: NSMI, 2.2 percentage points per month [95% posterior interval: 1.8, 2.6] SMI-LCT, 1.6 percentage points per month [1.3, 1.9]; SMI-LCI, 0.8 percentage points per month [0.4, 1.8]. The NSMI group, on average, had a steeper maximal growth rate than

Table 3. Model estimates and posterior intervals for key growth features.

		Population average		Estimated population	
Growth curve measure	**Estimate**	***Mdn***	**95% Interval**	***Mdn***	**95% Interval**
Intelligibility at age of 8 years (%)	NSMI	92	[88, 95]	93	[73, 98]
	SMI-LCT	82	[72, 89]	80	[3, 97]
	SMI-LCI	63	[43, 81]	52	[3, 96]
	NSMI – SMI-LCT	9.8	[3.3, 19.7]	12.0	[−13.6, 90.1]
	SMI-LCT – SMI-LCI	18.9	[−0.9, 39.5]	16.8	[−73.7, 86.9]
	NSMI – SMI-LCI	29.2	[11.1, 47.8]	38.9	[−7.5, 91.0]
Asymptote (%)	NSMI	93	[89, 95]	94	[75, 98]
	SMI-LCT	86	[74, 94]	86	[4, 100]
	SMI-LCI	74	[49, 90]	64	[3, 100]
	NSMI – SMI-LCT	6.3	[−0.7, 17.2]	6.1	[−17.3, 90.5]
	SMI-LCT – SMI-LCI	12.1	[−7.0, 37.0]	10.3	[−81.8, 91.9]
	NSMI – SMI-LCI	18.6	[3.5, 42.5]	27.8	[−12.4, 90.6]
Age of steepest growth (months)	NSMI	39	[36, 42]	39	[26, 52]
	SMI-LCT	54	[50, 59]	54	[33, 75]
	SMI-LCI	51	[34, 72]	51	[23, 82]
	NSMI – SMI-LCT	−15.4	[−21.2, −9.6]	−15.8	[−41.3, 9.9]
	SMI-LCT – SMI-LCI	3.1	[−17.9, 21.0]	3.33	[−34.7, 37.5]
	NSMI – SMI-LCI	−12.3	[−32.8, 4.9]	−12.4	[−45.4, 18.0]
Steepest growth rate (percentage points per month)	NSMI	2.2	[1.8, 2.6]	2.2	[1.2, 3.5]
	SMI-LCT	1.6	[1.3, 1.9]	1.5	[0.1, 2.9]
	SMI-LCI	0.8	[0.4, 1.8]	0.6	[0.0, 3.2]
	NSMI – SMI-LCT	0.56	[0.08, 1.04]	0.78	[−0.81, 2.60]
	SMI-LCT – SMI-LCI	0.77	[−0.17, 1.30]	0.70	[−1.80, 2.26]
	NSMI – SMI-LCI	1.32	[0.41, 1.88]	1.50	[−0.79, 2.94]

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

the SMI-LCT group, $slope_{NSMI} - slope_{SMI-LCT} = 0.6$ [0.1, 1.0]. Because the average growth rate for the SMI-LCI group had high uncertainty, the difference between the SMI-LCT and SMI-LCI group averages also had high uncertainty, $slope_{SMI-LCT} - slope_{SMI-LCI} = 0.8$ [−0.2, 1.3]. To aide interpretation, however, we stipulate that a group difference in growth rate greater than 0.25 percentage points per month is a *meaningful* difference. (In other words, differences of .25 or smaller are too small to be clinically meaningful.) The posterior probability that there was a meaningful difference between the SMI-LCT and SMI-LCI group averages, $P(slope_{SMI-LCT} - slope_{SMI-LCT}, > .25)$, was .90; equivalently, the posterior probability that these differences were clinically irrelevant was only .10.

The NSMI group had less between-children variability in the rate of maximum growth. The maximum growth rate in the NSMI group had a 95% prediction interval for a new, as-yet unobserved child of [1.2, 3.5] percentage points per month, compared to [0.1, 2.9] in the SMI-LCT group and [0.0, 3.2] in the SMI-LCI group. This finding is apparent in the population prediction bands for Figure 1 (fourth row). Some children in the SMI-LCT and SMI-LCI groups showed very little growth, and their maximum growth rates were close to zero. Thus, the population estimates for the groups had to allow for these kinds of growth trajectories.

Intelligibility at 8 Years of Age

The estimated average age-8 intelligibility for each group was NSMI, 92% [95% posterior interval: 88%, 95%] SMI-LCT, 82% [72%, 89%]; SMI-LCI, 63% [43%, 81%]. As with the midpoint feature, there was high uncertainty for the average in the SMI-LCI group. Nevertheless, the probability that the SMI-LCT group had higher average age-8 outcomes than the SMI-LCI group was .97.

There was much less variability in the between-children prediction for age-8 intelligibility in the NSMI group compared to the SMI-LCT and SMI-LCI groups. In Figure 1, in the observed data (first row) and in the between-children predictions (fourth row), the intelligibility scores for the NSMI group at 96 months are nearly all above 75%, whereas in the SMI-LCT group, the scores range from nearly 0% to nearly 100%. Specifically, the 95% population prediction intervals for the groups were as follows: NSMI [73%, 98%], SMI-LCT [3%, 97%], and SMI-LCT [3%, 96%]. Although they both covered the same range of outcomes, SMI-LCT outcomes were higher on average than SMI-LCI (based on the group differences reported above).

Question 2: How Does the Presence of Language Comprehension Impairment Influence Predicted Intelligibility Growth Trajectories for Children With SMI?

To address this question, we examine a hypothetical clinical scenario: Suppose we encounter a 4-year-old with SMI. What age-8 intelligibility outcomes are plausible, and how does the presence of language comprehension impairment change the distribution of plausible outcomes? To address these questions, we had our model simulate growth trajectories for a hypothetical, unobserved child from the SMI-LCT and SMI-LCI groups. We considered two scenarios: a child who is 40% intelligible at the age of 4 years and a child who is 3% intelligible at the age of 4 years. Figure 2 shows the distribution of predicted age-8 outcomes for each scenario by SLPG.

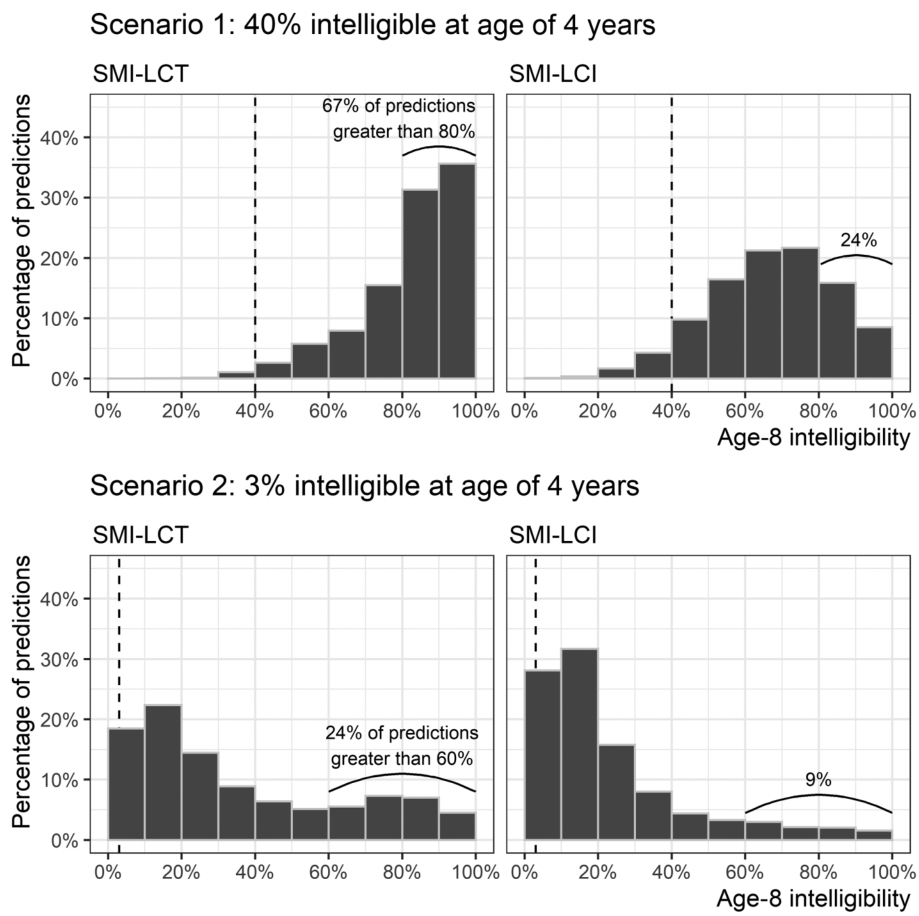
For Scenario 1 (40% intelligibility), if the hypothetical child with SMI had typical language comprehension at age of 4 years, then two thirds of predicted intelligibility outcomes resulted in intelligibility that was greater than 80%. If the child with SMI had impaired language comprehension, then only one quarter of outcomes resulted in intelligibility that was greater than 80%. The presence of SMI with typical language comprehension abilities predicted higher intelligibility outcomes with the most plausible band of outcomes in Figure 2 in the 90%–100% range. Moreover, impaired comprehension led to more uncertain predictions with three quarters of intelligibility predictions falling into the 50%–90% range. Thus, impaired language comprehension acted like a risk factor for intelligibility growth with more pessimistic and uncertain predictions.

For Scenario 2 (3% intelligibility), most outcomes were in the 0%–30% range for both the typical language comprehension group and the impaired language comprehension group. Thus, a hypothetical child with very low intelligibility at age of 4 years was not expected to demonstrate much growth by age of 8 years. In this scenario, the predictions for the SMI-LCT group showed more uncertainty. The predictions for the SMI-LCT child were bimodal: There was a (flatter, wider) second mode in the histogram around 80% intelligibility. Note that, in the *observed* data (as in Figure 1), there are roughly two sets of trajectories in the SMI-LCI group: Most children had age-8 intelligibility scores above 60%, but a few children show very limited growth and never exceed 40% intelligibility. In the forecasting, the model appears to be assigning the hypothetical child to one of these two trajectory sets (hence, two modes), and thus, one fourth of the model's predictions fell into the 60%–100% range. In the SMI-LCI case, the possibility (intelligibility above 60%) was very nearly ruled out with only one tenth of outcomes falling into the 60%–100% range. In this scenario, typical language comprehension leads to increased uncertainty by including more optimistic predictions for a child with very low intelligibility.

Discussion

In this study, we examined patterns of multiword intelligibility growth for children with CP in different speech language profile groups. We also examined how SMI—alone or in the presence of comorbid language comprehension impairment—impacted intelligibility growth. We discuss our results with emphasis on similarities and differences

Figure 2. Effect of language comprehension impairment in predicting intelligibility outcomes at age of 8 years in simulated children with speech motor impairment (SMI). Top row: Predicted outcomes for a 4-year-old with SMI and 40% intelligibility (vertical dashed line). The presence of language comprehension impairment (right) leads to more pessimistic, more variable predictions. Bottom row: Predicted outcomes for a 4-year-old with SMI and 3% intelligibility (vertical dashed line). The most likely outcome is very limited intelligibility growth, but the typical language comprehension (left) leads to greater uncertainty in the predicted outcomes with one-fourth of predictions falling above 60% intelligibility. NSMI = no speech motor impairment; SMI-LCT = speech motor impairment with typical language comprehension; SMI-LCI = speech motor impairment with impaired language comprehension.



between findings for the same children on single-word intelligibility development from our earlier work.

Question 1: Do Children With Different Speech-Language Profiles Show Different Patterns of Multiword Intelligibility Growth in Their Connected Speech? (Specifically, How Do Age of Steepest Growth, Rate of Steepest Growth, and Intelligibility at 8 Years of Age Vary Between Groups?)

We found that, on average, children without SMI (NSMI) showed earlier ages of steepest growth, approximately 1 year earlier than for children in the two SMI groups. Children in the NSMI group also showed steeper growth rates and better speech intelligibility outcomes compared to children in the SMI groups. In fact, the estimated intelligibility outcomes at age of 8 years for the NSMI group were, on average, 10 percentage points and 30 percentage points higher than the SMI-LCT group and the SMI-LCI group, respectively. Moreover, we found less between-children variation in the NSMI group; these children generally showed relatively homogenous growth trajectories and intelligibility outcomes. Although the absence of SMI at age of 4 years is predictive of favorable intelligibility outcomes for children with CP, our findings suggest that these children reach an average intelligibility of 92%, which is likely to be less intelligible than typically developing peers at age of 8 years. It is important to note that studies using methods comparable to the present one have not been conducted, and therefore, standards for intelligibility of typically developing children at 8 years of age are not available. This is an important direction for future research and has direct bearing on the interpretation of results from this study.

We also examined two other groups of children with SMI: One group had typical language comprehension (SMI-LCT), and one group had impaired language comprehension (SMI-LCI). These two groups showed more variability in their growth patterns and more uncertainty in estimated group averages than those in the NSMI group. That said, children in the SMI-LCT group had maximum growth rates that were, on average, twice as large as the rates for the SMI-LCI group. The SMI-LCT group also had estimated age-8 intelligibility outcomes that were approximately 20 percentage points higher than children in the SMI-LCI group. These findings are similar to our previous results examining single-word intelligibility, which suggested that outcomes were generally better for children who had SMI without concomitant language comprehension impairment (Hustad, Mahr, Broman, & Rathouz, 2020). It is important to note, however, that children in the SMI-LCI group were smallest in number and there is considerable variability in outcomes among all children with SMI. However, our results continue to support the idea that outcomes may be more consistent and more optimistic for children who do not have co-occurring language comprehension impairment.

We compared findings of this study on multiword intelligibility with findings from our earlier work examining growth of single-word intelligibility in the same children (Hustad, Mahr, Broman, & Rathouz, 2020), and the results of this study suggest some differences in the course of development for intelligibility of connected speech relative to the development of intelligibility for single-word productions. Specifically, findings of this study suggest that age of steepest growth may be later for multiword intelligibility than for single-word intelligibility for two of the groups: 4 months later for NSMI and 10 months later for SMI-LCT. Estimates were too uncertain for the SMI-LCI group; therefore, we refrain from making comparisons and generalizations about findings from this group. Rate of growth, however, was consistent within profile groups for single words and multiword utterances, across the two studies. These findings are consistent with what is known about important advancements that occur in children's speech and language abilities during the time frame of this study, most notably the ability to produce longer and more complex utterances from both a linguistic and speech motor control perspective. Finally, intelligibility outcomes at 8 years of age were considerably higher within each group for multiword intelligibility than for single-word intelligibility. Specifically, for both groups of children with SMI, intelligibility was about 20 percentage points higher at 8 years of age for multiword utterances than for single words. For children with NSMI, it was about 10 percentage points higher for multiword utterances than for single words. Again, this finding is consistent with the literature showing an intelligibility advantage for multiword utterances over single-word utterances as children get older (Hustad, Mahr, Natzke, & Rathouz, 2020)

Question 2: How Does the Presence of Language Comprehension Impairment Influence Predicted Intelligibility Growth Trajectories for Children With SMI?

To address this question beyond our initial conclusions addressed in Question 1, we performed two sets of simulations to look at the developmental implications of typical versus impaired language comprehension for children with SMI. For a child with 40% intelligibility at 4 years of age, a child with typical comprehension is most likely to be in the 80%–100% intelligibility range at 8 years of age, but if the child has language comprehension impairment instead of typical language, the most likely outcomes are lower and more uncertain (50%–90% range). In contrast, for a child with very low intelligibility, the most likely outcome is limited growth (less than 30% intelligibility). In this case, the predictions for a child with typical language comprehension include some more optimistic outcomes. In both sets of simulations, typical language comprehension led to higher predictions for intelligibility outcomes, and impaired language comprehension led to lower predictions. Between the two scenarios, however, the key predictor was age-4 intelligibility: Moderate to high growth levels were plausible with a starting point of 40% intelligibility, but not from a starting point of 3% intelligibility.

These results suggest the presence of language comprehension impairment along with SMI represents a sort of "two-hit" risk factor for intelligibility growth. That is, expected intelligibility outcomes at 8 years of age are diminished for children who have both SMI and language comprehension impairment relative to those who have SMI with typical language comprehension. This finding adds to a growing body of literature suggesting negative effects on speech motor control when the language system is taxed in children with language impairment (Saletta et al., 2018; Vuolo & Goffman, 2018). Another possibility is that language comprehension impairment indexes overall CP severity, so the addition of language comprehension impairment as a comorbidity signifies greater neurological involvement. Although we did not examine intellectual abilities among children in this study, our earlier work suggests that language comprehension impairment is highly correlated with the presence of intellectual disability (Soriano & Hustad, 2020). Recent studies provide clear evidence that children with CP and comorbid intellectual disability have different outcomes than those without comorbid intellectual disability (Sigurdardottir & Vik, 2011; Tan et al., 2020); thus, our findings are consistent with other recent studies. A key implication of this work is that early consideration of augmentative and alternative communication systems and strategies should be given to children with CP who have language comprehension impairment across all levels of severity to support speech development, language development, and functional communication development beyond the limits of their speech production.

Our results also have implications for monitoring early intelligibility development. Children in the NSMI group, on average, had passed their age of greatest growth by 48 months (the age when children were classified into profile groups). The average age of greatest growth for the SMI-LCT group, however, was somewhere between 49 and 59 months. These children were classified when their intelligibility was developing most rapidly. Early intervention focused on speech production for both groups of children with SMI beginning at or before age of 4 years could capitalize on this developmental momentum to help children make additional intelligibility gains. However, additional studies are needed to examine the potential benefits of timing intervention to coincide with developmental growth.

Limitations and Future Directions

There are several important limitations to this study. First, our sample of children with CP was drawn from the Upper Midwest region of the United States. The sample may not be fully representative of the overall population of children with CP because we were not able to recruit from the full pool of all children with CP due to privatized health care, research ethics board restrictions, and privacy laws in the United States. Replication of this work on a larger, more representative sample is necessary. That said, to our knowledge, this is one of the largest, longest running active cohort of children with CP who are being prospectively followed for direct measurement of speech and language development.

Although we had many observations over time on individual children, we had a relatively small number of participants in this study, and a very small proportion of these children had SMI-LCI. This small n and general heterogeneity among the children made group comparisons difficult and resulted in a considerable uncertainty with regard to outcomes for this group. In addition, over the course of this study, most children received speech and language therapy. As a result, we do not know the extent to which the observed growth was due to intervention, to development, or to some combination of the two. Treatment timing should be addressed in future studies that attempt to quantify intervention effects over time.

Some children show very limited growth in both of the SMI groups. These are the very flat lines in the first two rows of Figure 1 where four children in the SMI-LCT group and two children in the SMI-LCI group never cross 20% intelligibility after age of 84 months. These children have the lowest expected outcomes, so they make up an important subgroup for clinical decision making. Ideally, a classification system would be able to identify children in this subgroup as early as possible, and in this case, the three levels of the SLPG framework are not fine grained enough to highlight these highest risk children. That said, these children tend to have low intelligibility at younger ages, so they begin to diverge from their peers after age of 5 years.

The SLPG framework is one of several different classification schemes for considering communication in children with CP. This framework does not currently consider expressive language abilities because of measurement confounds with speech intelligibility deficits. As a result, conclusions regarding the impact of language on speech growth are limited to the receptive modality. Future research should examine language profiles more comprehensively to further explore the relationships between speech and language development in children with CP.

Within the SLPG framework, a high-risk subgroup appears in the SMI groups. It would be interesting to examine speech growth on the basis of different classification approaches such as the Viking Speech Scale (Pennington et al., 2013), the Functional Communication Classification System (Caynes et al., 2019), and the Communication Function Classification System (Hidecker et al., 2011). Future studies should examine how well these other systems predict speech outcomes, such as intelligibility. We provide descriptive information regarding the functional motor abilities of children with CP in this study; however, we did not examine gross motor functional classification level, manual ability classification level, or cognitive abilities in the context of speech intelligibility development or in the context of our SLPG classifications. These variables should be considered in future studies.

Our analysis framework only focused on intelligibility in connected speech, although we also measured single-word intelligibility for this sample (Hustad, Mahr, Broman, & Rathouz, 2020). A future direction, methodologically, would be to simultaneously study both intelligibility types and take advantage of the correlation between single-word and multiword intelligibility. One might ask, for example, whether predictions about age-8 outcomes are improved by knowing *both* intelligibility scores measured at age of 4 years.

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