

Longitudinal growth of receptive language in children with cerebral palsy between 18 months and 54 months of age

KATHERINE C HUSTAD^{1,2}  | ASHLEY SAKASH² | AIMEE TEO BROMAN³ | PAUL J RATHOUZ^{2,3}

1 Department of Communication Sciences and Disorders, University of Wisconsin – Madison, Madison, WI; **2** Waisman Center, University of Wisconsin – Madison, Madison, WI; **3** Department of Biostatistics and Medical Informatics, University of Wisconsin School of Medicine and Public Health, Madison, WI, USA.

Correspondence to Katherine C Hustad at Department of Communication Sciences and Disorders, University of Wisconsin – Madison, 1975 Willow Drive, Madison, WI 53705, USA.
E-mail: kchustad@wisc.edu

PUBLICATION DATA

Accepted for publication 21st March 2018.
Published online

ABBREVIATIONS

LCAE	Language comprehension age equivalency
NSMI	No speech motor involvement
ROC	Receiver operating characteristic
SMI	Speech motor involvement

AIM We examined receptive language developmental trajectories between 18 months and 54 months for three clinical speech-language profile groups of children with cerebral palsy (those with speech motor involvement, without speech motor involvement, and with anarthria) and quantified differences from age-level expectations. We identified latent classes of comprehension development, related these classes to clinical profile groups, and examined how well early receptive language predicted outcomes.

METHOD We used a prospective longitudinal design. Eighty-five children with cerebral palsy (43 females, 42 males) were followed longitudinally from 18 to 54 months of age. Children were seen two to eight times (322 data points). Children were classified into clinical profile groups. Language comprehension age-equivalent scores were the primary measures of interest.

RESULTS Children with anarthria had significant language delays, limited developmental change over time, and comprised their own latent class. Children with speech motor impairment had slight receptive language delays over time. Children with no speech motor impairment had age-appropriate receptive language over time. Early language comprehension scores were highly predictive of later latent profile group membership.

INTERPRETATION Early language comprehension abilities are highly predictive of language comprehension growth trajectory and suggest that children with early language delay, particularly those who are non-speaking, should receive language intervention to support development.

Many children with cerebral palsy (CP) experience some type of speech, language, and/or communication problem.^{1,2} Problems may include dysarthria, a speech motor disorder that often has critical detrimental impacts on speech intelligibility, language and/or cognitive difficulties, or a combination of both speech and language/cognitive problems.³ Any one or more of these can lead to functional communication challenges. In addition, gross and fine motor involvement affecting the limbs can have adverse effects on the ability to use gestures, pointing, and written modes of communication.

Recently, longitudinal work has shown that early speech production ability is highly predictive of later outcomes.⁴ Specifically, children with CP who speak well enough to repeat utterances in an elicitation task by 2 years of age make faster gains in intelligibility and utterance length, resulting in better speech production abilities at 4 years of age. This work also showed that children with CP who were not yet talking at 2 years of age were very likely to have significant speech, language, and communication

challenges at 4 years of age.⁴ However, parallel studies have not yet provided a detailed examination of trajectories of change in language development among these children.

Language abilities in children with CP can vary and seem to be related both to severity and type of motor involvement, as well as the presence of concurrent intellectual disabilities.^{5,6} Studies generally suggest that non-speaking children with severe motor impairment, regardless of type of motor disorder, have delayed language comprehension. Within motor involvement type, children with spastic CP tended to have language comprehension deficits that were more severe and generally below age expectations relative to language comprehension in children with dyskinetic CP.⁷ In addition, receptive communication growth trajectories seem to be more favorable for children with CP without intellectual disabilities.⁸ However, what growth trajectories look like over time and the extent to which rate of change follows developmental expectations is unknown. In addition, the speech motor abilities of children with CP in previous studies have not

been characterized; thus, a thorough understanding of how development of receptive language relates to speech production is not available.

In the present study, we sought to examine change in receptive language among children with CP on the basis of speech production abilities using a prospective longitudinal cohort design. A main goal was to examine developmental change in receptive language by speech-language profile group (those with speech motor involvement [SMI]; those with no speech motor involvement [NSMI]; and those with anarthria) following our earlier work,^{3,9} and to examine trajectories of development relative to age-level expectations. Specific questions for the present study were as follows. (1) What do receptive language developmental trajectories from 18 to 54 months of age look like for each of three clinical profile groups (which were determined at 48–54mo)? To what extent, and how, do children in each group differ from age-level expectations? (2) Are there distinct and discernable (latent) patterns of language development based on receptive language growth trajectories from 18 to 54 months of age across children with CP? How many different patterns or groups are present in the data, what do they look like, and how do they relate to clinical profile groups? (3) How well does early life language comprehension performance, at age 24 to 30 months, predict future language comprehension growth in terms of these latent patterns?

METHOD

Participants

Participants were selected from a larger cohort of children ($n=139$) participating in a longitudinal study on communication development in children with CP. Between 2005 and 2012, children were recruited through local and regional neurology and psychiatry clinics in the Upper Midwestern region of the USA. Children were between 18 months and 60 months old upon initial enrollment. Children were seen twice per year, roughly 6 months apart, until their eighth birthday, after which they were seen for yearly visits. Criteria for inclusion into the larger study required that children had a medical diagnosis of CP and hearing within normal limits as documented by either formal audiological evaluation or distortion-product otoacoustic emission screening. To be included in data presented in this paper, we required each child to have contributed at least two longitudinal time points and to have no codiagnosis of autism spectrum disorder. Ethical approval for this study was obtained from the University of Wisconsin – Madison Institutional Review Board for Social and Behavioral Sciences. All parents and children provided informed consent to participate in this study.

A total of 85 children (43 females, 42 males) met our criteria and were included in the data set presented here. Each child contributed two to eight data points, for a total of 322 data points across the 85 children, yielding a mean of 3.8 (SD 1.6) and median of four data points per participant. All children were from homes where English was the

What this paper adds

- There are two growth trajectories for language comprehension among children with cerebral palsy.
- Children with speech motor impairment had a constant 6-month receptive language delay.
- Children without speech motor impairment had age-appropriate receptive language.
- Non-speaking children had significant receptive language delay.
- Early language comprehension change was highly predictive of later trajectory group.

primary language. Children were born in the USA between 2001 and 2009. Demographic information including CP diagnosis is presented in Table I.

Materials and procedures

For the larger prospective longitudinal study, a speech-language evaluation protocol was administered by a research speech-language pathologist in a sound-attenuating suite. For the present study, receptive language results were of primary interest. The following measures were administered, depending on the child's age, developmental level, and motor skill profile: The Test of Auditory Comprehension of Language, Third Edition,¹⁰ the Preschool

Table I: Demographic characteristics of children by speech-language profile group

	No speech motor involvement at 48–54mo	Speech motor involvement at 48–54mo	Anarthria at 48–54mo	All children
Sex				
Male	14	12	16	42
Female	5	24	14	43
GMFCS level				
I	12	8	1	21
II	6	11	5	22
III	0	9	0	9
IV	1	8	6	15
V	0	0	18	18
Type of cerebral palsy				
Spastic				
Hemiplegia	11	13	1	25
Diplegia	5	6	3	14
Triplegia	0	1	0	1
Quadriplegia	0	7	18	25
Unknown	1	0	0	1
Dystonic	0	1	2	3
Choreo-athetotic	0	1	0	1
Ataxic	0	4	0	4
Mixed	1	0	4	5
Unknown	1	3	2	6
Vision				
Within normal limits	17	12	6	35
Corrected	1	19	6	26
Uncorrected	0	1	1	2
CVI	0	0	13	13
Other	1	4	4	9
LCAE (mo)	52.8	43.9	9.7	

CVI, cortical visual impairment; GMFCS, Gross Motor Function Classification System; LCAE, language comprehension age equivalency.

Language Scale, Fourth Edition,¹¹ and the Peabody Picture Vocabulary Test, Fourth Edition.¹²

Children younger than 36 months received the Preschool Language Scale, which assesses earlier acquired skills with portions administered by parent interview. For all children 36 months of age and older, the Test of Auditory Comprehension of Language was attempted. An important advantage of this test is that completion requires limited motor skills, as response options are presented in a field of three discrete pictures which can be selected using manual direct selection or partner-directed scanning. However, if a child was not able to participate in administration of the Test of Auditory Comprehension of Language, the Preschool Language Scale was administered. The Peabody Picture Vocabulary Test was administered to children who could not tolerate the longer Test of Auditory Comprehension of Language, but who could participate in picture pointing tasks by manual direct selection or partner-directed scanning. On a child-by-child and item-by-item basis, standard administration procedures were adapted to enable participation in testing for items involving manual manipulation. Instructions in the technical manuals were followed for setting up adaptations. Scores on each of these three language tests were used interchangeably on the basis of well-established content validity for the measurement of receptive language.^{10,11} We note that the Peabody Picture Vocabulary Test only provides information about receptive vocabulary and thus is not a comprehensive measure of receptive language abilities. However, because of its ease of administration and frequent use in hard-to-test populations, we included it to provide at least a cursory window into language comprehension for those children who could not complete other tests.

Scores on the three different language tests were converted to age-equivalent values following the technical manual for each of the tests. This placed all scores on the same age-based scale, regardless of the test, and allowed us to examine whether average age-equivalent comprehension score was equal to chronological age. It also allowed us to test statistically whether trajectories of change followed a slope of 1, which would be expected under a typical developmental trajectory (e.g. age-equivalent language comprehension at age 36mo is expected to equal 36mo).

At 48 to 54 months, children were classified into one of three speech-language profile groups following our earlier work.^{3,9} We used a retrospective classification approach where we looked backwards in time at early behavior, given knowledge of later outcomes because our studies suggest that children with CP cannot be readily classified using the speech-language profile group paradigm before 4 years of age.¹³ Studies seeking to identify earlier emerging classification paradigms using prospective modelling are currently underway, but until results are available, findings from retrospective classification models provide useful information about how children get to their later classification outcomes.

Classifications were made on the basis of clinical judgement of the presence or absence of speech motor impairment and the presence or absence of language impairment. Children who were classified as having NSMI had no clinical evidence of speech or language impairment based on clinician observation during the data collection session and were confirmed by review of video and audio recordings after the session.

Children who were classified as having SMI, by definition, had clinical evidence of dysarthria. Speech motor impairment was determined through clinical observation of the presence or absence of common features of dysarthria, including facial asymmetry, drooling, hypernasality, short breath groups, breathy, harsh, or wet vocal quality, imprecise articulation, and consonant or vowel substitutions, distortions, or omissions that were not age-appropriate. Perceptual judgements were made during two tasks: (1) a delayed imitation task in which the child produced a standard set of sentences ranging from two to seven words in length following an adult model;¹⁴ and (2) a spontaneous speech sample either between the child and parent or the child and clinician. Receptive language abilities at 48 to 54 months of age were determined on the basis of standardized test results for the measure that each child was able to complete at 48 to 54 months as described above. Determination of receptive language delay was based on standard scores that were 1.5 standard deviations below age expectations per the technical manual of respective tests.^{10–12} Children who had language comprehension impairment and SMI were classified as SMI-LCI; those who had language comprehension that was typically developing and SMI were classified as SMI-LCT.

Children who were classified as having anarthria produced fewer than five words or word approximations on the basis of parent report and clinician observation during the data collection session. Classification into one of these four groups was done by a research speech-language pathologist on the basis of objective testing data (i.e. language comprehension scores, intelligibility scores, oral motor performance) and on review of videotaped spontaneous communication samples, as needed. See Table I for demographic characteristics of children by speech-language profile group.

Statistical analyses

To address the first research question, language comprehension age equivalencies (LCAEs) were modelled against chronological age in months in a repeated-measures linear mixed model with random effects for each participant's intercept and slope, fitting separate models for each profile group. For each profile group, we simultaneously tested whether the mean LCAE at 24 months and at 48 months was different from 24 and 48 respectively, and whether the average slope (of LCAE vs age) for each clinical profile group was different from 1. We note that, within this model, the mean LCAE value of 24 at 24 months or of 48 at 48 months, and a slope of 1, represent the 2 degrees of

freedom joint null hypothesis that the group was developing typically (i.e. along a line with intercept 0 and slope 1). When we rejected this null hypothesis, we then went on to separate post hoc tests of whether the mean LCAE at 24 months and 48 months differed from 24 months and 48 months respectively, and/or the slope differed from 1.

To address the second research question, we performed growth mixture model analysis of the LCAE trajectories across 18 to 54 months. In these models, we assumed that the population was divided into some number of latent classes, each characterized by a specific typical growth trajectory (an intercept and a slope), and modelled by a class-specific linear mixed model.¹⁵ The growth mixture model adjusted for correlation between repeated measures on the same participant, and allowed for multiple latent classes, which were assigned to participants on the basis of the posterior probability of being in that class. Our model specification included random effects for each participant's intercept and slope, and allowed for class-specific variances and covariances of the random effects, as well as class-specific error variance. The models were fitted with an extended Marquardt algorithm, which is a variant of Newton–Raphson optimization.^{16,17} A set of 100 random initial values was used to avoid optimizing the likelihood at local maxima.^{18,19} The Bayesian information criterion was used for model selection.²⁰ We assumed there were one to three underlying latent classes. We described the fitted classes using the estimated mean trajectory, coplotted with individual observed trajectories, color-coded according to their posterior class assignment.

To address the third research question, we exploited the fitted growth mixture model with two classes. In the context of this model, we examined how well language comprehension measured only at 24 months and 30 months using early language comprehension tests described above predicted latent class membership (and, by extension, future language comprehension growth) across the entire 18-month to 54-month age span. To quantify the prediction, we performed a receiver operating characteristic (ROC) curve analysis²¹ for predicting membership in one of the two latent classes as a function only of measurements at 24 months and 30 months, obtaining both the ROC curve and the area under the curve. We executed this computationally using a large-scale simulation model. We used the estimated latent class model parameters to simulate measures at 24 months and 30 months for 1000 computer-generated ‘children’, half with underlying class 1, and half with class 2. Simulated measures were random values generated from a bivariate normal distribution using the estimated mean and covariance for each latent class at 24 months and 30 months. Then, for each simulated child, we estimated a risk score: the likelihood of being in class 1 versus class 2. Finally, we estimated the diagnostic value of the risk score in predicting class 2 versus class 1 membership in an ROC analysis. Bootstrap resampling,²² from our original sample of 85 children, was used to obtain standard errors and to construct confidence intervals for

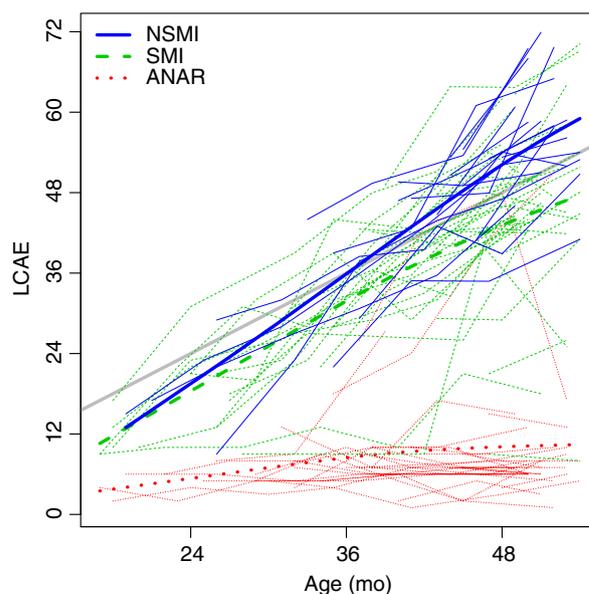


Figure 1: Language comprehension age-equivalent (LCAE) scores over time by speech-language profile group. The gray line represents expected growth trajectories for typically developing children. NSMI, no speech motor impairment profile group; SMI, speech motor impairment profile group (pooled across language abilities); ANAR, anarthria profile group. [Color figure can be viewed at wileyonlinelibrary.com].

model parameters, including the ROC curve and corresponding area under the curve.

RESULTS

Developmental trajectories by clinical profile group

Individual trajectories of LCAE scores are shown in Figure 1, by clinical profile group. Note that the gray line showing a slope of 1 in the figure reflects the expected trajectory for a typically developing child. A cubic smoothing spline with 3 degrees of freedom shows the average trajectory of LCAE versus chronological age for each clinical profile group.

Table II shows estimates from the repeated-measures random effects models for each of three clinical profile groups, and these are displayed in Figure 1 along with the individual trajectories for each participant. From the fitted models, we computed the estimated mean LCAE value for each group at ages 24 months and 48 months. These ages were chosen because they encompassed the largest number of observations while still representing the range of participant ages. In all three groups, the joint 2 degrees of freedom test of whether the mean at age 48 months differed from 48 and the slope differed from 1 was rejected ($p < 0.001$), so individual tests of age-specific means and slopes were also performed (see Table II).

For the children who were unable to produce speech (anarthria group), at age 24 months and 48 months, the average LCAE was 6.1 (standard error 0.9) and

Table II: Random effects models of language comprehension age equivalency (LCAE) as a function of age, by speech-language profile group

	Coefficient	Standard error	LCL	UCL	Test	df	χ^2	$p(>\chi^2)$
ANAR: mean LCAE at 24mo	6.114	0.947	4.258	7.971	Mean LCAE at 24=24	1	356.6	$p<0.001$
ANAR: mean LCAE at 48mo	10.041	1.597	6.910	13.172	Mean LCAE at 48=48	1	564.7	$p<0.001$
ANAR: slope	0.164	0.068	0.030	0.297	Slope=1	1	151.0	$p<0.001$
ANAR					Mean LCAE at 48=48 and slope=1	2	739.1	$p<0.001$
SMI: mean LCAE at 24mo	17.647	1.339	15.023	20.271	Mean LCAE at 24=24	1	22.5	$p<0.001$
SMI: mean LCAE at 48mo	42.078	2.231	37.706	46.450	Mean LCAE at 48=48	1	7.0	0.008
SMI: slope	1.018	0.069	0.883	1.153	Slope=1	1	0.1	0.795
SMI					Mean LCAE at 48=48 and slope=1	2	23.1	$p<0.001$
NSMI: mean LCAE at 24mo	21.205	1.914	17.453	24.956	Mean LCAE at 24=24	1	2.1	0.144
NSMI: mean LCAE at 48mo	52.544	1.680	49.251	55.837	Mean LCAE at 48=48	1	7.3	0.007
NSMI: slope	1.306	0.089	1.132	1.480	Slope=1	1	11.9	$p<0.001$
NSMI					Mean LCAE at 48=48 and slope=1	2	13.1	0.001

ANAR, anarthria profile group; df, degrees of freedom; LCL, lower confidence limit; NSMI, no speech motor impairment profile group; SMI, speech motor impairment profile group (pooled across language abilities); UCL, upper confidence limit.

10.0 months (standard error 1.6) respectively. Both means were statistically lower than the chronological age of 24 months and 48 months ($p<0.001$). The slope was 0.16 (standard error 0.07), which was significantly different from 1 ($p<0.001$), reflecting growth that was considerably slower over time than developmental expectations.

For the SMI group, at age 24 months and 48 months, the average LCAE was 17.6 (standard error 1.3) and 42 months standard error 2.2) respectively. Both means were significantly lower than the chronological age of 24 months and 48 months ($p<0.001$) but much more in line with norms than the anarthria group. The slope of 1.02 (standard error 0.07) was not statistically different from 1 ($p=0.8$), reflecting a developmentally appropriate growth rate over time.

Among the NSMI group, at age 24 months and 48 months the average LCAE was 21.2 (standard error 1.9) and 52.5 months (standard error 1.7). While the former was not different from 24 ($p=0.14$), the latter was significantly higher than 48 ($p=0.007$). The slope was 1.31 months (standard error 0.099), which was significantly different from 1 ($p<0.001$), reflecting moderately accelerated growth (i.e. the slope of change over time was faster than expected for typically developing children) over this age range.

Patterns (latent classes) of language comprehension development

We fitted models with two and with three latent classes in the growth mixture modelling, and allowed the initial values to vary so as to find the highest likelihood, and not merely local maxima. The best model with two latent classes had a Bayesian information criterion of 2175.7, with an estimated 29 participants in class 1 and 56 participants in class 2, while the best model with three latent classes had a Bayesian information criterion of 2171.6 with three participants in class 1, 53 participants in class 2, and 29 participants in class 3. Note that descriptive examination of the data revealed that the three children who constituted class 1 in this model all had LCAEs that

got worse over time; one was in the anarthria group and two were in the SMI group. Even though the Bayesian information criterion was lower (indicating a better model fit) for the three-class solution, the very small class 1 detracted from model parsimony to a degree that all further analyses were done using the model with two latent classes. The estimated probability of class membership was 34% (95% CI 25–45) for class 1 and 66% (95% CI 55–75) for class 2.

LCAE growth trajectories for each participant are shown in Figure 2, which was obtained by randomly assigning each participant to a class on the basis of their posterior

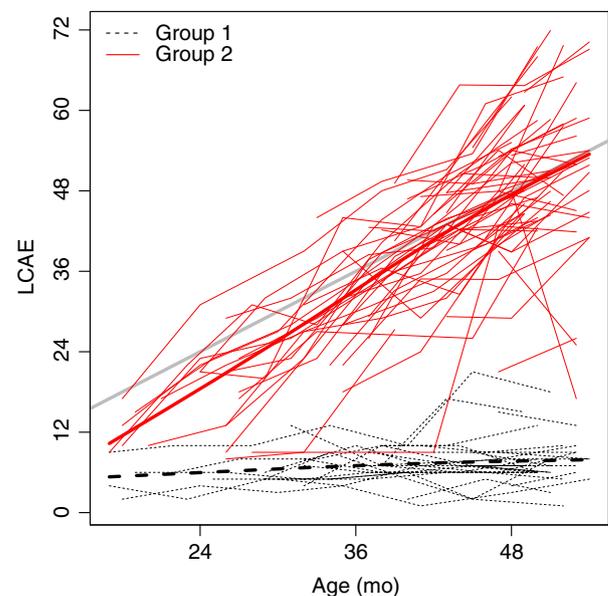


Figure 2: Language comprehension age-equivalent (LCAE) scores over time by latent class group. Group 1, developmentally delayed trajectory group; Group 2, developmentally typical trajectory group. The gray line represents expected growth trajectories for typically developing children. [Color figure can be viewed at wileyonlinelibrary.com.]

Table III: Latent class model with class-specific random effects models of language comprehension age equivalency (LCAE) as a function of age

	Coefficient	Standard error	LCL	UCL	Test	df	χ^2	$p(>\chi^2)$
Class 1: mean LCAE at 24mo	6.128	1.000	4.168	8.087	Mean LCAE at 24 class 1=24	1	319.6	$p<0.001$
Class 1: mean LCAE at 48mo	7.711	0.620	6.495	8.927	Mean LCAE at 48 class 1=48	1	4215.9	$p<0.001$
Class 1: slope	0.066	0.048	-0.028	0.160	Slope class 1=1	1	379.8	$p<0.001$
Class 1: Developmentally delayed group					Mean LCAE at 48 class 1=48 and slope class 1=1	2	4431.7	$p<0.001$
Class 2: mean LCAE at 24mo	19.639	1.367	16.960	22.318	Mean LCAE at 24 class 2=24	1	10.2	0.001
Class 2: mean LCAE at 48mo	47.450	1.168	45.160	49.740	Mean LCAE at 48 class 2=48	1	0.2	0.638
Class 2: slope	1.159	0.038	1.084	1.234	Slope class 2=1	1	17.2	$p<0.001$
Class 2: Developmentally typical group					Mean LCAE at 48 class 2=48 and slope class 2=1	2	18.5	$p<0.001$

Probability of latent class membership: Class 1=0.34 (CI=0.25, 0.45); Class 2=0.66 (CI=0.55, 0.75). Intercepts are in months.

probabilities of latent class membership. Note that posterior probabilities reflect the likelihood of being placed into each latent class, given observed data from the latent class model. We also include the linear mixed model fit of the average LCAE for each latent class. The figure shows clear separation in the two latent classes. Table III shows the estimates from the latent class models. In both classes, the joint test of whether the mean at age 48 months differed from 48 and the slope differed from 1 was rejected ($p<0.001$), so the individual tests of intercepts and slopes were also performed (see Table III).

Class 1 will hereafter be referred to as the ‘developmentally delayed trajectory’ group. For children in this group at age 24 months and 48 months, the average LCAE was 6.1 (standard error 1.0) and 7.7 months (standard error 0.62) respectively. Both means were statistically lower than the chronological age of 24 months and 48 months ($p<0.001$). The slope was 0.07 (standard error 0.05), and was significantly different from 1 ($p<0.001$), reflecting growth that was considerably slower over time than developmental expectations.

Class 2 will hereafter be referred to as the ‘developmentally typical trajectory’ group. For children in the developmentally typical trajectory group, at age 24 months and 48 months, the average LCAE was 19.6 (standard error 1.4) and 47.4 months (standard error 1.2) respectively. While the former was significantly different from 24 ($p=0.001$), the latter was not different from 48 ($p=0.6$). The slope for class 2 was 1.16 (standard error 0.04), which was close to but statistically different from 1 ($p<0.001$), reflecting growth that was slightly accelerated relative to developmental expectations.

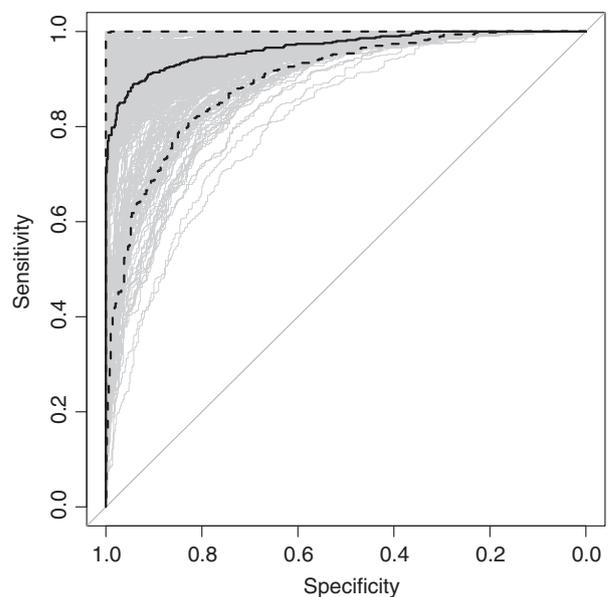
Table IV: Speech group versus latent classes

	Class 1 (%)	Class 2 (%)
ANAR	27 (93.1)	3 (5.4)
SMI	2 (6.9)	34 (60.7)
NSMI	0 (0)	19 (33.9)

Fisher’s exact test for count data, $p<2.2\times 10^{-16}$. ANAR, anarthria profile group; NSMI, no speech motor impairment profile group; SMI, speech motor impairment profile group (pooled across language abilities).

To investigate the degree to which the latent trajectory classes predicted clinical profile group at 54 months, we cross-classified the two on the basis of the aforementioned random class assignment (Table IV). Nearly all (93.1%) children in the developmentally delayed trajectory group were classified into the anarthria profile group. Of the two children who were classified into the developmentally delayed trajectory group, but who were members of the SMI profile group, both made very little change in their LCAE scores, and both had very limited speech production ability that was primarily at the single-word production level with limited intelligibility.

Nearly all (94.6%) of the children in the developmentally typical trajectory group were classified in either the SMI or NSMI groups. All the three children who were in the typical trajectory group, but who were members of the anarthria group, made considerable change over time in their language comprehension scores, similar to the

**Figure 3:** Receiver operating characteristic for simulated risk score, with bootstrapped confidence interval.

children in the SMI group with whom they were classified. This strong separation of categories provides prospective validation of our data-derived latent classes.

Prediction of later language comprehension growth from early performance

On the basis of our fitted latent class model, we estimated the strength of predication of latent class membership using only data that would be available at 24 months and 30 months. The ROC curve for diagnostic ability of the resulting 24-month to 30-month risk score in classifying participants into class 1 or 2 only on the basis of these two early data points is shown in Figure 3, along with the bootstrap distribution of ROC curves (providing a visual display of the statistical variation) and a 95% confidence band of ROC curves (also expressing statistical variation). The estimated area under the ROC curve was 0.97 (95% bootstrap confidence 0.90–1.0), suggesting that early performance across two time points provides very strong separation between the two latent classes of language comprehension growth in children with CP.

DISCUSSION

This study had three key findings: (1) there were two separate latent class growth trajectories for language comprehension development among children with CP; (2) the two growth trajectories closely mapped onto clinical profile groups such that one trajectory group comprised all but three children in the anarthria group who were unable to speak at 48 to 54 months and the other group comprised all but two children who were able to speak at 48 to 54 months (regardless of whether speech motor impairment was present); (3) early language comprehension change was highly predictive of later trajectory group membership. Results are discussed in the context of the binary latent classes, which were closely tied to speech production ability, and their associated clinical profiles groups.

Children who were not able to speak and were classified clinically as having anarthria at 48 to 54 months generally had significant language comprehension delays early in life and limited developmental change over time. Cross classification of children in the anarthria group with latent class findings based on growth trajectories revealed that 93% of what we refer to as the developmentally delayed trajectory latent class comprised children with anarthria at 48 to 54 months of age. The receptive language delay for this group was considerable, both in terms of absolute difference relative to age expectations at 24 months and 48 months and rate of change over time. Early language comprehension scores for children who could not speak were highly predictive of later latent profile group membership on the basis of our latent class analyses. Inspection of Figure 2 shows that all but a very few children in the developmentally delayed trajectory group had findings consistent with mean group results.

Previous research has demonstrated that children who were not able to speak and who had severe gross motor involvement also tended to have significant language

comprehension delays.^{6,8} In the present study, most of the children in the anarthria group had severe gross motor involvement (Gross Motor Function Classification System [GMFCS] levels IV and V), and most children had spastic CP; thus our findings, both in terms of the population and their language abilities, are consistent with other literature. It is important to note, however, that information on manual ability such as that provided by the Manual Ability Classification System²³ has the potential to refine our understanding of the extent to which manual ability may have interfered with testing procedures. Similarly, new tools exist for measuring language comprehension in individuals with severe motor challenges; however, they are not yet available in the English language.^{24,25}

While the findings of the present study may suggest a poor outlook for children with anarthria, there are several critical considerations that bear discussion. First, although we sought to reduce the impacts of motor impairment on the measurement of language comprehension, some level of motor activity was necessary for children to be given credit for language comprehension items above about a 12-month developmental level. Although we were able to provide modifications, some children with CP still had difficulty using a motor modality, particularly those in GMFCS level V. Given this testing limitation, a key conclusion is that barriers to language comprehension measurement are persistent and show little change between 18 months and 54 months of age. This finding is supported by research examining the stability of functional gross motor skills as captured by the GMFCS.²⁶ Specifically, children's GMFCS levels tend to be very stable with growth, indicating that children do not outgrow their gross motor challenges with development. On the whole, many of these children simply cannot demonstrate language comprehension abilities through standardized tests using the necessary motor movements.

Children who had CP and were able to speak at 48 to 54 months comprised two groups: those with SMI and those with NSMI. Collectively, these two clinical groups of children comprised one latent class on the basis of LCAE growth trajectories. Cross classification of children in the SMI and NSMI groups with latent class findings showed that 95% of children in this latent class were in one of these two clinical profile groups, with a developmentally appropriate growth trajectory. This combined group had typical receptive language both in terms of mean LCAE scores at 24 months and 48 months and in terms of change over time, although change was slightly accelerated relative to typical expectations. Early language comprehension scores from children who were able to speak at 48 to 54 months were highly predictive of later latent profile group membership on the basis of our observed latent class analyses.

When examined as individual clinical profile groups, children who could speak at 48 to 54 months, but who had SMI, tended to have a constant receptive language delay of about 6 months that persisted through the developmental age range of this study (54mo); however, their rate of

change over time was developmentally appropriate, such that a 1-month gain in chronological age yielded a 1-month gain in LCAE. The second group of children who could speak at 48 to 54 months, those with NSMI, had age-appropriate receptive language early in life with accelerated growth over time, resulting in language comprehension abilities slightly above age expectations at 48 months. Previous studies of children with NSMI have suggested that they may have reduced intelligibility relative to typically developing peers.²⁷ Findings of the present study suggest there may be a gap between speech development, which seems to lag, and language comprehension development, which is slightly accelerated.

Limitations and future directions

This study provides a preliminary glimpse into the complex relationships between speech and language development; however, there were several important limitations to this work. The question of how gross motor skills affected each child's ability to demonstrate language comprehension is an important one. We suggest that results for children in the anarthria group may be more a reflection of persistent motor barriers to testing than language comprehension development, which highlights the value of further development and advancement of tools to measure language comprehension in individuals with severe motor impairment. Also critical is the development of alternative access approaches that are more effective for demonstrating receptive language. Accurate characterization of and models for receptive language acquisition are critical for developing augmentative and alternative communication interventions that enhance language

acquisition and allow children to meet their underlying language potential.

Results of this study showed that early language comprehension change is highly predictive of growth trajectory to 54 months of age. This finding has important clinical implications, suggesting that reduced language comprehension performance at ages as young as 24 months should be a rapid trigger for early intervention. Specifically, aggressive intervention should be considered to enhance access to alternative modalities for both expressive and receptive language, including augmentative and alternative communication approaches such as aided language stimulation and alternative access tools.^{28,29}

In this study, we used standardized receptive language assessment measures that focus on identifying underlying deficits. These measures do not inform our understanding of functional receptive language skills in context, which may be different. Future studies should seek to examine the development of functional comprehension skills in a dyadic communicative context in order to inform how well children with CP are able to participate in real-world communication situations.

ACKNOWLEDGEMENTS

This study was funded by grant R01DC009411 from the National Institute on Deafness and Other Communication Disorders, National Institutes of Health. Support was also provided by a core grant to the Waisman Center, U54 HD090256, from the National Institute of Child Health and Human Development, National Institutes of Health. The authors have no financial relationships relevant to this article to disclose. The authors have stated that they had no interests that could be perceived as posing a conflict or bias.

REFERENCES

1. Bax M, Tydeman C, Flodmark O. Clinical and MRI correlates of cerebral palsy: the European cerebral palsy study. *JAMA* 2006; **296**: 1602–8.
2. Nordberg A, Miniscalco C, Lohmander A, Himmelmann K. Speech problems affect more than one in two children with cerebral palsy: Swedish population-based study. *Acta Paediatr* 2013; **102**: 161–6.
3. Hustad KC, Gorton K, Lee J. Classification of speech and language profiles in 4-year-old children with cerebral palsy: a prospective preliminary study. *J Speech Lang Hear Res* 2010; **53**: 1496–513.
4. Hustad KC, Allison KM, Sakash A, McFadd E, Broman AT, Rathouz PJ. Longitudinal development of communication in children with cerebral palsy between 24 and 53 months: predicting speech outcomes. *Dev Neurorehabil* 2017; **20**: 323–30.
5. Pirila S, van der Meere J, Pentikainen T, et al. Language and motor speech skills in children with cerebral palsy. *J Commun Disord* 2007; **40**: 116–28.
6. Mei C, Reilly S, Reddihough D, Mensah F, Pennington L, Morgan A. Language outcomes of children with cerebral palsy aged 5 years and 6 years: a population-based study. *Dev Med Child Neurol* 2015; **58**: 605–11.
7. Geytenbeek JJ, Vermeulen RJ, Becher JG, Oostrom KJ. Comprehension of spoken language in non-speaking children with severe cerebral palsy: an explorative study on associations with motor type and disabilities. *Dev Med Child Neurol* 2015; **57**: 294–300.
8. Vos RC, Dallmeijer AJ, Verhoef M, et al. Developmental trajectories of receptive and expressive communication in children and young adults with cerebral palsy. *Dev Med Child Neurol* 2014; **56**: 951–9.
9. Hustad KC, Oakes A, McFadd E, Allison KM. Alignment of classification paradigms for communication abilities in children with cerebral palsy. *Dev Med Child Neurol* 2016; **58**: 597–604.
10. Carrow-Woolfolk E. Test for Auditory Comprehension of Language. 3rd edn. Austin, TX: Pro-Ed, 1999.
11. Zimmerman I, Steiner V, Pond R. Preschool Language Scale-4. 4th edn. San Antonio, TX: Psychological Corporation, 2002.
12. Dunn LM, Dunn DM. Peabody Picture Vocabulary Test. 4th edn. San Antonio, TX: Psychological Corporation, 2007.
13. Hustad KC, Allison K, McFadd E, Riehle K. Speech and language development in 2-year old children with cerebral palsy. *Dev Neurorehabil* 2014; **17**: 167–75.
14. Hodge M, Daniels J. TOCS+ Intelligibility Measures. Edmonton, AB: University of Alberta, 2007.
15. Proust-Lima C, Philipps V, Liqueur B. Estimation of extended mixed models using latent classes and latent processes: the R package lcmdm. *J Stat Softw* 2017; **78**: 1–56.
16. Marquardt DW. An algorithm for least-squares estimation of nonlinear parameters. *J Soc Ind Appl Math* 1963; **11**: 431–41.
17. Proust C, Jacqmin-Gadda H. Estimation of linear mixed models with a mixture of distribution for the random effects. *Comput Methods Programs Biomed* 2005; **78**: 165–73.
18. Biernacki C, Chrétien S. Degeneracy in the maximum likelihood estimation of univariate Gaussian mixtures with EM. *Stat Probab Lett* 2003; **61**: 373–82.
19. Biernacki C, Celeux G, Govaert G. Choosing starting values for the EM algorithm for getting the highest likelihood in multivariate Gaussian mixture models. *Comput Stat Data Anal* 2003; **41**: 561–75.
20. Raftery A. Bayesian model selection in social research. *Sociol Methodol* 1995; **25**: 111–63.
21. Pepe MS. The Statistical Evaluation of Medical Tests for Classification and Prediction. Oxford: Oxford University Press, 2003.

22. Efron B, Tibshirani RJ. *An Introduction to the Bootstrap*. New York, NY: CRC Press, 1994.
23. Eliasson AC, Krumlinde-Sundholm L, Rosblad B, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Dev Med Child Neurol* 2006; **48**: 549–54.
24. Geytenbeek JJ, Heim MM, Vermeulen RJ, Oostrom KJ. Assessing comprehension of spoken language in non-speaking children with cerebral palsy: application of a newly developed computer-based instrument. *Augment Altern Commun* 2010; **26**: 97–107.
25. Geytenbeek JJ, Mokkink LB, Knol DL, Vermeulen RJ, Oostrom KJ. Reliability and validity of the C-BiLLT: a new instrument to assess comprehension of spoken language in young children with cerebral palsy and complex communication needs. *Augment Altern Commun* 2014; **30**: 252–66.
26. Wood E, Rosenbaum P. The gross motor classification system for cerebral palsy: a study of reliability and stability over time. *Dev Med Child Neurol* 2000; **42**: 292–6.
27. Hustad KC, Schueler B, Schultz L, DuHadway C. Intelligibility of 4-year-old children with and without cerebral palsy. *J Speech Lang Hear Res* 2012; **55**: 1177–89.
28. Ronski M, Sevcik RA, Barton-Hulsey A, Whitmore AS. Early intervention and AAC: what a difference 30 years makes. *Augment Altern Commun* 2015; **31**: 181–202.
29. Drager K, Light J, McNaughton D. Effects of AAC interventions on communication and language for young children with complex communication needs. *J Pediatr Rehabil Med* 2010; **3**: 303–10.

RESUMEN**CRECIMIENTO LONGITUDINAL DEL LENGUAJE RECEPTIVO EN NIÑOS CON PARÁLISIS CEREBRAL ENTRE LOS 18 Y 54 MESES DE EDAD**

OBJETIVO Examinamos las trayectorias del desarrollo del lenguaje receptivo de tres grupos clínicos entre 18 y 54 meses, según el perfil del habla y el lenguaje de niños con parálisis cerebral (aquellos con afectación motora, sin afectación motora del habla y con anartria) y diferencias cuantificadas de las expectativas del nivel de edad. Identificamos tipos de desarrollo de comprensión latentes, relacionando estas clases con grupos de perfiles clínicos y examinamos qué tan bien pronosticaron los resultados del lenguaje receptivo temprano.

METODO Usamos un diseño longitudinal prospectivo. Ochenta y cinco niños con parálisis cerebral (43 mujeres, 42 varones) fueron seguidos longitudinalmente desde los 18 a los 54 meses de edad. Los niños fueron vistos de dos a ocho veces (322 puntos de datos). Los niños fueron clasificados en grupos de perfiles clínicos. Los puntajes de la comprensión del lenguaje equivalentes a la edad fueron las principales medidas de interés.

RESULTADOS Los niños con anartria tuvieron retrasos significativos en el lenguaje, con un cambio limitado en el desarrollo a lo largo del tiempo y conformaron su propio tipo latente. Los niños con discapacidad motora del habla tuvieron retrasos leves en el lenguaje receptivo al pasar el tiempo. Los niños sin discapacidad motora del habla tenían un lenguaje receptivo apropiado para su edad a lo largo del tiempo. Las puntuaciones de comprensión del lenguaje tempranas fueron altamente predictivas de la membresía al grupo latente según el perfil posterior.

INTERPRETACION Las habilidades tempranas de comprensión del lenguaje son altamente predictivas de la trayectoria de crecimiento de la comprensión del lenguaje y sugieren que los niños con retraso temprano del lenguaje, particularmente aquellos que no hablan, deben recibir intervención del lenguaje para apoyar el desarrollo.

RESUMO**DESENVOLVIMENTO LONGITUDINAL DA LINGUAGEM RECEPTIVA EM CRIANÇAS COM PARALISIA CEREBRAL ENTRE 18 E 54 MESES DE IDADE**

OBJETIVO Nós examinamos as trajetórias do desenvolvimento da linguagem receptiva entre 18 e 54 meses para grupos de crianças com paralisia cerebral com três perfis clínicos de linguagem receptiva (aqueles com envolvimento motor da fala, sem envolvimento motor da fala, e com anartria) Identificamos as classes latentes de desenvolvimento da compreensão, relacionadas a estes perfis clínicos, e examinamos quão bem a linguagem receptiva precoce foi preditiva dos resultados.

MÉTODO Usamos um desenho prospectivo longitudinal. Oitenta e cinco crianças com paralisia cerebral (43 do sexo feminino, 42 do sexo masculino) foram acompanhadas longitudinalmente dos 18 aos 54 meses de idade. As crianças foram vistas de duas a oito vezes (322 pontos de dados) e foram classificadas nos grupos de acordo com o perfil clínico. Os escores equivalentes à idade para compreensão da linguagem foram as medidas primárias de interesse.

RESULTADOS Crianças com anartria tinham atraso significativo na linguagem, mudança desenvolvimental limitada com o passar do tempo, e formaram sua própria classe latente. Crianças com envolvimento motor da fala tinha leve atraso na linguagem receptiva com o tempo. Crianças sem comprometimento motor da fala tinham linguagem receptiva adequada para a idade com o passar do tempo. Os escores precoces de compreensão da fala foram altamente preditivos do pertencimento ao grupo de perfil latente posteriormente.

INTERPRETAÇÃO Habilidades precoces de compreensão da linguagem são altamente preditivas da trajetória de desenvolvimento da compreensão da linguagem, e sugerem que crianças com atraso precoce, particularmente as que não falam, devem receber intervenção de linguagem para favorecer seu desenvolvimento.