

Longitudinal Development of Receptive Vocabulary in Children with Cerebral Palsy and Anarthria: Use of the MacArthur-Bates CDI

Michael Molinaro, Aimee Teo Broman, Paul J. Rathouz & Katherine C. Hustad

To cite this article: Michael Molinaro, Aimee Teo Broman, Paul J. Rathouz & Katherine C. Hustad (2019): Longitudinal Development of Receptive Vocabulary in Children with Cerebral Palsy and Anarthria: Use of the MacArthur-Bates CDI, Developmental Neurorehabilitation, DOI: [10.1080/17518423.2019.1646829](https://doi.org/10.1080/17518423.2019.1646829)

To link to this article: <https://doi.org/10.1080/17518423.2019.1646829>



Published online: 29 Jul 2019.



Submit your article to this journal [↗](#)



View Crossmark data [↗](#)



Longitudinal Development of Receptive Vocabulary in Children with Cerebral Palsy and Anarthria: Use of the MacArthur-Bates CDI

Michael Molinaro^a, Aimee Teo Broman^b, Paul J. Rathouz^c, and Katherine C. Hustad^a

^aUniversity of Wisconsin, Madison, WI, USA; ^bUniversity of Wisconsin School of Medicine and Public Health, Madison, WI, USA; ^cUniversity of Texas, Austin, TX, USA

ABSTRACT

Objective: To examine receptive language growth in children with cerebral palsy (CP) and anarthria using a parent-reported measure of vocabulary.

Method: Scores from 47 children (29 males) with CP and anarthria were obtained from the vocabulary checklists on the MacArthur-Bates Communication Development Inventories (MCDI) and analyzed to examine the distribution of receptive language growth. Linear trajectories of word composite scores were created using a linear-mixed model, incorporating between two and ten data points per child.

Results: Three different growth trajectories emerged: approximately 23% grew by 100 or more words per year, 13% grew by 50–100 words per year, and 64% grew by 50 words per year or less. Age-four vocabulary was strongly correlated with rate of increase in vocabulary.

Conclusion: Receptive vocabulary scores from the MCDI are increasing at a reduced pace for most children with CP and anarthria. More sensitive measures of language assessment are necessary to gain a complete picture of their language ability levels.

ARTICLE HISTORY

Received February 12, 2019

Revised July 17, 2019

Accepted July 18, 2019

KEYWORDS

Cerebral palsy; anarthria;
receptive language;
longitudinal design;
intellectual disability

Introduction

Cerebral Palsy (CP) is defined as a group of non-progressive motor impairments which result from an injury to the brain during the early stages of life. Due to the nature of the disorder, assessment and treatment often primarily focuses on the resulting physical limitations and their impact on the early stages of development. However, population-based studies have drawn increased awareness to co-occurring deficits areas. A multitude of factors, including CP type, time of onset, and cortical areas affected, result in a wide range of cognitive, motor, and communication abilities in these children.¹ One study showed that over 60% of children diagnosed with CP had some type of communication impairment.² Communication deficits can include speech motor impairments such as dysarthria, which frequently manifests as speech that sounds slurred, slow, and difficult to understand. In addition, many children with CP may have receptive and/or expressive language impairment. Receptive language impairments affect the ability to understand words and sentences. Expressive language impairments affect the ability to produce words and sentences that are age appropriate in length and complexity, as well as productive vocabulary. Production of expressive language is often hindered by dysarthria, but deficits can often be compensated for through the use of alternative expressive communication modalities such as augmentative and alternative communication (AAC) systems and strategies. Research focused on specific aspects of speech and/or language suggests that communication delays at 2 years of age may affect considerably more children with CP than earlier estimates have suggested.³

Several classification systems have been designed to reduce heterogeneity and identify common features among sub-groups of children with CP. Classification systems for individuals with CP exist for domains including: gross motor skills,⁴ manual abilities,⁵ feeding,⁶ and communication.^{7–10} One such classification model, proposed by Hustad et al.⁹ subdivides children based on two axes, speech motor abilities, and language abilities. Speech-language profile groups (SLPGs) from this model are: (1) children with CP who have no speech motor impairment (NSMI), (2) children with CP who have speech motor impairment (dysarthria) and typical language comprehension (SMI-LCT), (3) children with CP who have speech motor impairment (dysarthria) and language comprehension impairment (SMI-LCI), and (4) children with CP who have speech motor impairment that is so severe that they are unable to produce functional speech (anarthria). Using this paradigm, growth trajectories of individual profile groups over time have been examined.^{3,11,12} One recent study examined communication development between 24 and 53 months and found that early communication abilities were highly predictive of later profile group membership.¹¹ For example, children with CP who were not producing speech by 24 months were very likely to be classified as anarthric or as having speech motor impairment at 53 months. Estimates vary regarding how many children diagnosed with CP could also be considered anarthric. At five to 6 years of age, Mei and colleagues¹³ reported that 24% of their population-based sample was unable to speak. Other studies have suggested that this

number could be higher – up to 44%.³ The present study seeks to examine receptive vocabulary change as reported by parents longitudinally from 17 to 83 months of age among children with CP who have anarthria and are thus unable to speak.

Children who have CP and anarthria have received little research attention, despite the fact that they comprise a significant portion of the population. These children frequently present with severe gross and fine motor impairment that affects their ability to gesture, point, or manipulate objects to demonstrate knowledge. For decades, researchers and clinicians have been aware of testing barriers for this population via existing behavioral measures.¹⁴ In spite of technological advances, these barriers persist and options are very limited for measuring language ability in children who are unable to speak.¹⁵ Existing language assessments require the ability to talk, the fine motor control to manipulate objects and/or point, and a certain degree of limb control for motor movements such as pointing.

One recent study examined longitudinal development of receptive language in children with CP.¹² Results from standardized receptive language measures showed that children with anarthria demonstrated limited growth between the ages of 18 and 54 months, and often maintained very low language comprehension scores over time. However, it is difficult to determine if these results are indicative of actual language abilities or are instead a reflection of motor impairment that prohibits the ability to demonstrate underlying language skills. Studies examining the Gross Motor Function Classification System (GMFCS)⁴ in children with CP have established a correlation between the severity of gross motor impairment and severity of communication deficits.¹⁶ In particular, children with the most severe gross motor involvement also tend to have the most severe communication challenges. The speech production aspect of communication can be readily observed in children with CP and quantified in terms of severity (or inability to produce speech). However, language is difficult to accurately measure in the presence of severe motor involvement and severe dysarthria/anarthria, which limit the use of testing modalities including speech, and manual movement such as pointing and manipulating objects. Thus, quantifying latent abilities such as receptive language is challenging. These factors have important implications for intervention, particularly for the use of augmentative and alternative communication (AAC) systems. In one study, Griffiths & Addison¹⁷ noted that 100% of children classified as GMFCS levels IV or V had expressive and/or receptive communication impairments which required the use of AAC systems. A precise understanding of a child's language abilities is an important step in matching a child to an appropriate AAC system. Additionally, a growing body of work has shown that AAC systems are not only used as a means of expressive output but can serve a prominent role in providing language input.¹⁸

Alternative measures of receptive language abilities suggest promising potential. Geytenbeek, Heim, Vermeulen, & Oostrom,¹⁹ developed an assessment called the Computer-Based Instrument for Low Motor Language Testing (C-BiLLT), which was designed to bypass challenges presented by motor limitations through use of alternative access modalities such as eye gaze. A recent cross-sectional study using

the C-BiLLT found that non-speaking children with CP were still improving their receptive language scores at age 12.²⁰ Given these findings, the study of additional means for capturing language comprehension abilities of non-speaking children with CP is an important area for exploration.

Indirect measures of receptive language abilities are a simple avenue for uncovering underlying ability levels that could inform AAC interventions. Parent-report measures, such as the MacArthur-Bates Communicative Development Inventories,²¹ are well-established tools for measuring language growth in children with and without disabilities.^{22,23} The MCDI has been used to predict language abilities in preterm, late-talkers, and language-impaired children.^{23–25} More recently, the MCDI has been used to examine language growth in children with developmental disabilities, including: autism spectrum disorders (ASD), down syndrome, intellectual disability, and cochlear implant recipients.^{26–30} Several of these studies have examined language growth well beyond the intended 30-month age range^{25,29} and found that the MCDI continued to be a useful measure of vocabulary development. This measure has enabled researchers to get below the detection limits of standardized language tests when assessing children with more complex communication needs. However, the MCDI has not been used in studies examining communication abilities in children with CP. The MCDI could provide a more sensitive index of receptive vocabulary and its change over time for children with limited speaking and motor abilities relative to traditional measurements involving behavioral responses.

In the present study, we sought to examine how receptive vocabulary as reported by parents on the MCDI changes longitudinally between the ages of 17–83 months among children with CP who are unable to speak. We were interested in the rate of vocabulary growth as rate of language growth has been suggested to be an indicator of learning efficiency in children³¹ and could potentially serve as a rationale for provision of increased intervention services. Here we provide the first analysis examining the slope in words per year of vocabulary growth in children with CP. We also examined vocabulary size at 4 years, an age when children are entering formal education (preschool or 4k programs in the US) and is thus a critical time for communication development. For children with CP, 4 years of age is particularly germane because studies in our lab have suggested that most children with CP who will acquire the ability to produce spoken words do so by this time³² and because this was the age point from which we identified the presence of anarthria in our sample. We addressed the following specific questions:

- (1) How do children who are unable to speak grow with regard to receptive vocabulary? Specifically, what is the slope of their growth, as well as the range and distribution of trajectories among children?
- (2) What is the range of children's receptive vocabulary size at the midpoint of our longitudinal timeframe (48 months) and how does it relate to growth?

Given the heterogeneity and complexity of the population, we expect that parent-report measures of receptive vocabulary via the MCDI will show receptive vocabulary acquisition to have

considerable variability in this sample such that some children will demonstrate growth that is more accelerated than others. We also predict that a child's receptive vocabulary knowledge at 48 months will be indicative of their estimated growth. This hypothesis was founded on the finding that the fourth year of life seems to be a time when disability profiles in children with CP tend to become relatively stable.³²

Materials and Methods

Participants

Participants were selected from a larger cohort of children with a clinically confirmed diagnosis of cerebral palsy (CP; $n = 139$) who were enrolled in a prospective longitudinal study on communication development. In the present study, we examined only children who were clinically classified as anarthric, which we operationally defined as those who were unable to produce more than five words or word approximations using speech, per clinical observation and parent report at 48 months of age. Additional inclusion criteria for the present study required each child to have hearing within normal limits and to have contributed at least two longitudinal data points between the ages of 17 and 83 months. A total of 47 children (29 males, 18 females) met these requirements and were included in this paper. Table 1 shows demographic data including: CP type, Gross Motor Function Classification System (GMFCS)⁴ level, Manual Ability Classification System (MACS)⁵ level, and vision status. All participants came from

homes where English was the primary language. Each child contributed between two and ten data points, for a total of 289 data points across the 47 children. The mean number of data points per child was 6.13 (SD 2.15) and the median was seven data points per child. Table 2 displays the distribution of data points by age group in yearly intervals. Table 3 describes the participants use of an AAC system during the course of their participation in the study. Participants were separated into one of four categories: children who used an AAC system less than 25% of the time ($n = 21$), children who used an AAC system for 25–49% of the time ($n = 7$), children who used an AAC system for 50–74% of the time ($n = 6$), and children who used an AAC system for 75% or more of the time ($n = 13$).

Materials and Procedures

The MacArthur Communication Development Inventories (MCDI) are well-established parent-report measures of early language abilities. These communication assessments are normed and validated on typical children between the ages of eight and 30 months.^{21,24,33} Test-re-test reliability is reported in the technical manual for the MCDI to be between .8 and .9. Validity has been well established; in particular, studies of concurrent validity and predictive validity as reported in the technical manual yield positive findings.²¹ The MCDI has been used extensively in language development research including studies of typically developing children and in children with developmental disabilities.^{23–30} We highlight that in the present study, we extend the use of the MCDI beyond the normative range of typical development for which the measures were created. While there are clear limitations to this, there is precedent in the literature where the MCDI has been used in older children with intellectual and developmental disabilities.^{31,34,35}

The MCDI comprises two separate forms. The first MCDI form, Words & Gestures (MCDI-WG; created for children eight to 16 months) includes two parts: early words & actions and gestures. For this study, we focused on part one, early words, and specifically the vocabulary checklist. The vocabulary checklist queries a child's understanding and production of 396 words that fall into 19 semantic categories (e.g. animals, vehicles, toys, foods, body parts). The vocabulary checklist is completed by a parent who checks one of two possible

Table 1. Demographic and clinical characteristics of anarthric children with CP.

Anarthric children ($n = 47$)	
Sex	
Male	29
Female	18
Type of CP	
Spastic	
Diplegia	3
Hemiplegia (left)	1
Hemiplegia (right)	0
Triplegia	0
Quadriplegia	20
Unknown	1
Hypotonic	4
Mixed	4
Dyskinetic	1
Unknown	13
GMFCS at 4 years	
I	0
II	4
III	2
IV	12
V	29
MACS at 4 years	
I	1
II	5
III	2
IV	27
V	12
Vision	
Within normal limits	11
Corrected	8
Uncorrected	3
Cortical Visual Impairment	21
Other	4

*GMFCS = Gross Motor Function Classification System;

*MACS = Manual Abilities Classification System.

Table 2. Number of total observations by age group for MCDI.

Months CA	12–23	24–35	36–47	48–59	60–71	72–83
Total observations	6	29	52	79	72	51

Table 3. Use of an AAC system at the time of each visit per parent report ($n = 47$ children).

	Number of children	% ($n = 47$)
Children who used an AAC system less than 25% of the time.	21	44.7%
Children who used an AAC system for 25–49% of the time.	7	14.9%
Children who used an AAC system for 50–74% of the time.	6	12.8%
Children who used an AAC system for 75% or more of the time.	13	27.7%

boxes for each individual word, indicating if their child understands the word or understands and says the word. Words in the checklist are left blank if the parent does not think his/her child understands them. For this population of anarthric children, we were only interested in the number of words the child understood. If a child understood every word on the MCDI-WG vocabulary checklist, they would receive a maximum score of 396.

The second MCDI form, Words & Sentences (MCDI-WS; created for children 17–30 months), follows the first form in a sequential fashion and also contains two sections: the words children use and sentences and grammar. Again, we only focused on part one which is comprised entirely of a 680-item vocabulary checklist. These words, separated into 22 semantic categories, queried a child's ability to produce the words verbally. We modified the published version of the MCDI-WS form to include an option for parents to mark that their child understands the word to capture receptive vocabulary data from children with anarthria. Parents completed the modified checklist for words and sentences in the same fashion as described for the words and gestures checklist. If a child understood every word on the MCDI-WS vocabulary checklist, they would receive a maximum score of 680. Note, however, that all of the 396 words on the MCDI-WG vocabulary checklist are also included on the MCDI-WS vocabulary checklist with some slight changes to the semantic categories to which each item was assigned.

As part of our experimental protocol, we sent MCDI forms to parents 14 days before each longitudinal visit. Parents first received the MCDI-WG form before their initial visit. After the first visit, selection and distribution of the specific form across children and visits were determined by a research speech-language pathologist who considered MCDI performance from the previous visit in determining which form to send. Generally, as a child's chronological age increased and a child's raw score on the MCDI-WG vocabulary checklist increased to above 100 (out of 396) parents were given the MCDI-WS form to avoid the possibility of ceiling effects. Parents returned the MCDI at the time of each visit.

Statistical Analysis

Analyses of the longitudinal receptive vocabulary data from the MCDI required us to address several issues. First, the age range of visits varied somewhat from child to child. Second, the number of visits was variable across children, and the visits were irregularly spaced for some children. Third, even while generally following a smooth trajectory of growth, the within-child variability of measures around that trajectory was substantial. For these three reasons, it was challenging to fit an individual growth curve separately to each child's data. To address these concerns, we estimated linear trajectories of MCDI scores using a linear-mixed model, regressing score on age, accounting for repeated measures on each child, and allowing for random intercept and slope for each child. Linear mixed models not only capture the average intercept and slope across the population of children but provide posterior, 'best', predictions of the child-specific intercept and slope for each child depending on the data available. For example, a child with only a few visits at young ages may yield moderate information on the intercept but weak

information on slope. The statistical model automatically borrows information from the population to make the best estimate of both the intercept and the slope for that child. For another child with several visits evenly spread out across the age range, less population information will be needed because data will be more informative about both intercept and slope. This feature of the linear mixed modeling approach accommodates missing data arising from variability in the number of visits across children and the irregular spacing of visits for some children.

To begin our analyses, we plotted raw MCDI scores against age in months for each child and overlaid the best fit linear trajectory for each child. We ordered the children by slope using a color gradient. We then plotted all of the raw data onto a single plot using this color gradient so that the population distribution of trajectories could be visually displayed. Finally, we plotted best fits of child-specific slopes versus best fits of scores at 48 months and examined the distributions of both 48-month scores and slopes in order to provide a succinct but comprehensive description of the distribution of trajectories experienced by this population.

Results presented in this paper are descriptive in nature, based on results of the fitted linear mixed model regression analyses described above. Because these data have never been examined in this manner, we did not perform inferential tests on these results.

Results

Descriptive results are shown in [Figures 1 and 2](#). Summary statistics, shown in [Figure 1](#) suggest that overall, children showed a gradual increase in MCDI scores over time, particularly from 18 to 53 months. These data also show the considerable variability within each age band. [Figure 2](#) shows individual growth trajectories, highlighting the variability among individual children. This variability is not unexpected given the complex communication ability profiles of children with anarthria, potentially ranging from severe intellectual disability to mild or no intellectual disability, all with the common feature of an inability to produce speech.

- (1) How do children who are unable to speak grow with regard to receptive vocabulary? Specifically, what is the slope of their growth, as well as the range and distribution of trajectories among children?

[Figure 2](#) shows observed trajectories of each of the 47 children who contributed two or more longitudinal data points to the data set. Data indicate that there are a wide range of growth trajectories for children with anarthria, with some (in red) not progressing at all and others (in purple and dark blue) showing dramatic improvements, especially in the three to five-year age range. In [Figure 3](#), the histogram of slopes (seen on the y-axis of the figure) shows that over 35% of children have very low or zero slopes, indicating little or no vocabulary acquisition over time, whereas a few children are estimated to increase their vocabulary by as much as 150 words/year. For example, the three children with the fastest growing vocabularies have slopes of 155.5 words/year, 157.6 words/year, and 175.8 words/year. Although the sample size is not

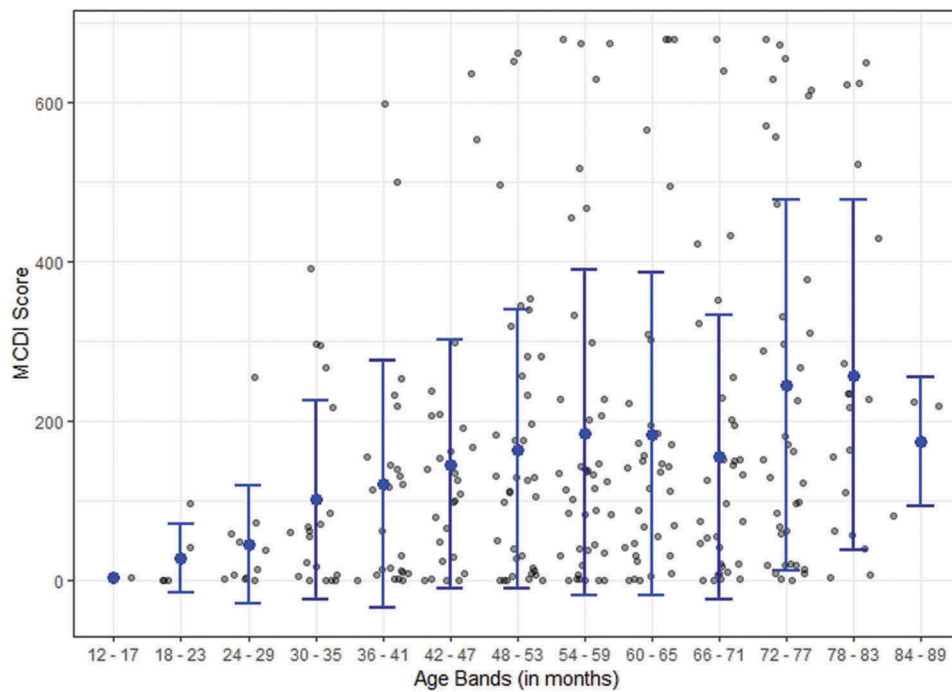


Figure 1. Raw scores on the MCDI receptive vocabulary checklist by 6-month age band. Individual data are plotted in gray, blue dots represent means, upper and lower error bars show standard deviations.

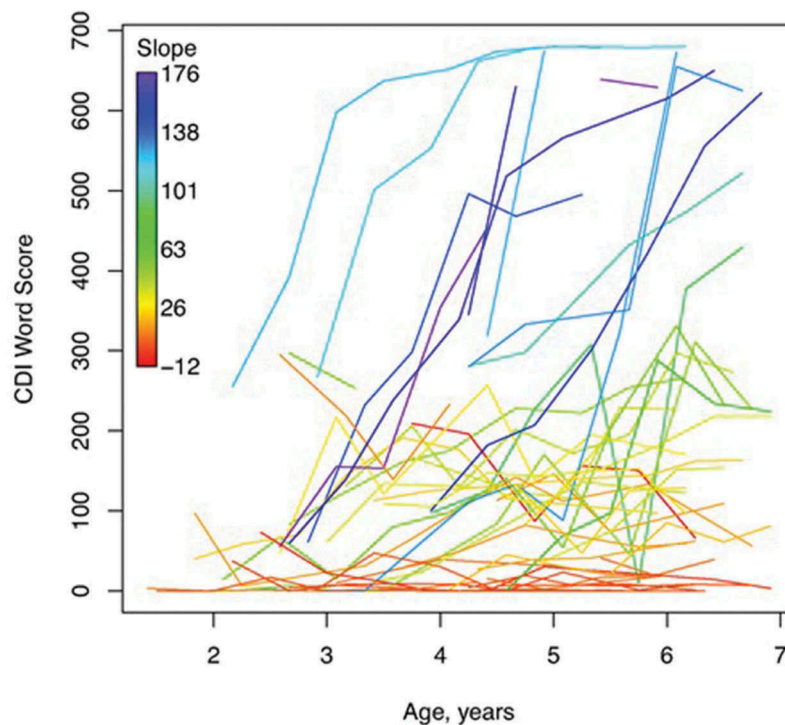


Figure 2. Individual receptive vocabulary growth on the MCDI from two to 7 years of age for children with anarthria. Note that slope is expressed in words per year. Lines represent individual children. Lines are colored according to the slope of growth in words per year.

large enough to confirm this feature, there is a suggestion of bimodality or trimodality, with a minority of children (approximately 23%) experiencing robust growth of over 100 words/year, the majority (64%) experiencing 50 words/year or less, and the few remaining children (13%) growing between 50 and 100 words per year.

- (2) What is the range of children's vocabulary size at 48 months (midpoint of the age span) and how does it relate to their estimated growth?

Figure 3 shows the distributions of estimated vocabulary at 4 years based on the fitted linear mixed model regression.

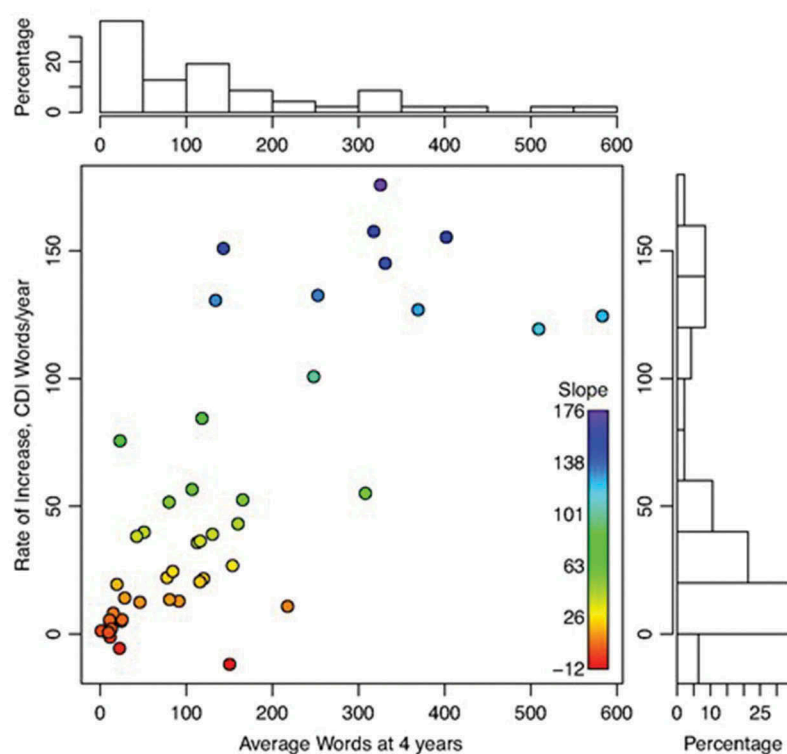


Figure 3. The scatter plot shows modeled receptive vocabulary size at 48 months by rate of increase (slope) in words per year. Each point represents an individual child and is colored according to the rate of growth in words per year. The histogram on the x-axis shows the distribution of vocabulary sizes at 48 months; while the histogram on the y-axis shows the distribution of children's rate of word growth per year.

Results indicate that vocabulary at 4 years is strongly correlated with slope (corr. = 0.77, 95% CI = [0.61, 0.86], $p < .0001$), such that children who are acquiring vocabulary more quickly have larger vocabularies at 4 years of age, which can be seen in the scatterplot. The histogram on the x-axis of Figure 3 shows that about 35% of children have 50 words or less in their vocabulary at age four, about 35% of children have between 51 and 200 words, and the remaining 30% of children have vocabularies as high as 500–600 words at age four on the MCDI. The highest vocabulary size at 4 years of age was 583 words.

Discussion

This study sought to examine how receptive vocabulary grew over time from 17 to 83 months in a group of 47 non-speaking children with cerebral palsy. We used an adapted version of the MCDI vocabulary checklists to quantify the range and distribution of receptive vocabulary growth as measured by parent report. Additionally, we were interested in children's receptive vocabulary size at 48 months, and how it related to children's vocabulary growth as they began their school-aged years. There were two main findings for this study. First, three groups of children emerged with regard to rate of growth. Second, the size of children's vocabulary at 48 months was a strong indicator of how quickly they continued to learn new words. The main findings of this study are discussed below.

Differences in the Rate of Vocabulary Growth

Previous literature has shown that receptive language growth in early childhood is limited for many children with CP and anarthria. Often, these children remain near or at the floor when standardized language assessments are used to quantify developmental change.¹² However, standardized language assessment tools require the ability to speak, point, gesture, and/or manipulate objects to demonstrate understanding. Geytenbeek and colleagues¹⁵ have argued that there are no adequate standardized language assessments which overcome the persistent motor impairment seen in the population of non-speaking children with CP. In the current study, using a parent-reported measure of receptive vocabulary, children in our sample showed gains in receptive vocabulary over time. Figure 1 displays group averages demonstrating slow but consistent receptive vocabulary growth from 17 to 83 months. While there is considerable variability between participants, overall, data indicate that children are learning new words each year. These findings support the notion that severe speech and motor impairments may mask a child's ability to demonstrate language growth on standardized assessments and that most children with anarthria are acquiring new receptive vocabulary through the elementary school years, and beyond.

Descriptive analyses of the patterns observed in growth trajectories among children with anarthria suggest that there may be up to three main groups of children: those learning over 100 words per year (23%; 11 children); those learning 50–100 words per year (13%; six children); and those learning

Table 4. Number of children by sex, type of CP, GMFCS score, mean vocabulary size at 48 months, and use of AAC, and rate of receptive vocabulary growth.

	< 50 words/ year	50–100 words/ year	> 100 words/ year
Number of children	30	6	11
Males; females	21; 9	4; 2	7; 4
Type of CP			
Spastic	16	4	5
Dyskinetic	0	0	1
Hypotonic	3	1	0
Mixed	2	0	1
Unknown	9	1	3
GMFCS at 48 mos.			
I	0	0	0
II	3	1	0
III	1	0	1
IV	8	0	4
V	18	5	6
Vocabulary size at 48 mos.			
Mean			
SD	65.4	133.8	329.0
Min	59.0	97.0	137.6
Max	1.5	23.4	134.3
	217.8	308.2	583.5
Use of AAC			
< 25% of sessions	16	4	1
25–49% of sessions	4	1	2
50–74% of sessions	3	0	3
≥75% of sessions	7	1	5

fewer than 50 words per year (64%; 30 children). Descriptive analyses of children in each of the aforementioned growth groups (see Table 4) show that children with the slowest growth (less than 50 words per year) had the smallest receptive vocabularies at age 4. The majority of these children had very limited access to AAC interventions, had severe motor impairment as indicated by GMFCS scores, and had primarily spastic CP. This was the largest group of children in the sample. Children who showed growth between 50 and 100 words per year were the smallest group and looked indistinguishable in terms of demographic variables from those children in the slowest growth group on all indices except vocabulary size at 4 years. Vocabulary size was nearly double that of the slowest growth group, although within group variability was considerably larger. Finally, children who showed vocabulary growth of more than 100 words per year looked very similar to the other two groups in terms of type of CP, severity of motor involvement, and male:female ratio. However, in this group, the majority of the children (72%) had access to AAC intervention for half or more of the sessions for which data were collected. From these data, it is not possible to make assumptions regarding causality. That is, we do not know if AAC use caused receptive vocabulary growth, or if better receptive vocabulary skills prompted service providers and families to pursue AAC. Although the absolute number of words children acquired in this study was small relative to typical expectations, receptive vocabulary growth of 50, 100, or 150 words per year could have an important impact on functional communication in the presence of appropriate AAC systems and services to support language and to engage in social interaction.^{17,18}

Our results reveal some parallels to previous literature suggesting there may be potential differences in cognitive profiles among children with CP and anarthria,³⁶ which may result in different rates of vocabulary acquisition. We did not

examine cognitive ability profiles in the present study; however, this variable is likely an important one that would shed light on language acquisition patterns observed in children with CP.

It is important to consider the findings of the present study in the context of the reporting tool parents used. The MCDI vocabulary checklists include a finite number of words that are known to be understood and used by typical children from eight to 30 months of age. We examined receptive vocabulary growth up to 83 months, thus these checklists may not adequately capture each child's entire receptive vocabulary. For example, children participate in home, school, and leisure experiences which provide exposure to many additional words that are not queried on the MCDI. Our findings are most likely a modest projection of receptive vocabulary growth in children with anarthria. While these findings provide useful information, they are not sufficient to ensure a comprehensive characterization of receptive abilities in this population. Tools such as the C-BiLLT^{37,38}, which was recently translated to English, offer promise for direct assessment of language comprehension. There are many advantages to direct assessment over indirect assessment via the MCDI. In particular, direct assessment allows for observation of language understanding via some output modality such as eye gaze, pointing, or scanning, leading to stronger evidence of comprehension than indirect measures based on parent impressions.

Predictive Nature of 48-Month Receptive Vocabulary Size

Research on typical language development has shown that word comprehension tends to precede word production.³⁹ This developmental sequence often remains true for children with developmental disabilities.⁴⁰ Normative data on typical language development provide context for considering receptive vocabulary in children with anarthria in this paper. An open repository for MCDI data, Wordbank, provides expressive language growth curves for typically developing children up to 30 months of age. According to Wordbank, a typically developing 30 month-old in the 50th percentile would have an expressive vocabulary of 541 words while the same aged child in the 75th percentile would have an expressive vocabulary of 613 words.⁴¹ Because comprehension tends to precede production in word learning, we can assume that a typical 30-month-old child in the 50th percentile understands at least 541 words and in the 75th percentile at least 613 words on the MCDI. However, at the age of 48–53 months, children with anarthria had a mean receptive vocabulary of 164.19 words (SD 174.86), well below typical peers who are 2 years younger. This is not surprising given the severity of the deficits and the complexity of communication challenges in children with anarthria, but the magnitude of this gap is noteworthy and once again highlights the need for interventions to foster language learning in children with anarthria.

When we investigated the association between a child's vocabulary size at 48 months and the rate of receptive vocabulary growth, results showed a strong correlation. This finding indicates that larger vocabulary size at 48 months is strongly associated with steeper receptive vocabulary growth over time, thus children with larger vocabularies at 48 months grow more quickly than those with smaller receptive vocabularies at 48 months. This result is

generally consistent with studies of typically developing children demonstrating that early language abilities (and particularly vocabulary) are highly predictive of growth⁴² and lends support to the argument that early language intervention is critical to maximizing later outcomes. For children with CP, this finding has important clinical implications for those with both smaller and larger receptive vocabularies. For example, all children may benefit from early intensive language intervention, particularly interventions focused AAC strategies such as aided language stimulation.^{43,44} Aided language stimulation is an intervention approach that provides explicit exposure to word learning through the use of multiple communication tools and modalities including spoken language, aided symbols, and referential pointing. This technique provides redundant exposure to new words within their natural context and has been shown to facilitate new word acquisition.^{45,46} For children with smaller vocabularies, aided language stimulation could target high impact core vocabulary. These high frequency and early developing words help provide a strong foundation for functional communication in a variety of contexts. For those children with more robust vocabularies at 48 months, aided language stimulation could focus on expanding higher level core words as well as fringe vocabulary (individualized, topic specific, or educational content words) in order to expand expressive language.

Limitations and Future Directions

There are several important limitations to the present study. First, while the sample size is considerable for this type of research, it is likely too small to be fully representative of the larger population of children with CP who have anarthria. Second, there may be limitations with parent-reported measures, as some parents may overestimate vocabulary while others may underestimate their child's vocabulary knowledge. Future studies should consider investigating alternative ways to capture language comprehension abilities, for example, using eye gaze methodologies. Third, language development is a complex and multifaceted process. This paper only examined receptive vocabulary; there are many other components of language comprehension (e.g. syntax, morphology, pragmatics) that should be considered in order to fully understand a child's language comprehension abilities. Lastly, we collected only cursory data on the type of AAC intervention each child received. We were not able to gather specifics about the child's AAC system, the available vocabulary, or whether the AAC system went back and forth from home to school. All these factors could significantly impact receptive language growth. Additionally, an examination of the type/frequency of the AAC intervention the children received was outside the scope of this study. A recent literature review shows that intervention with aided AAC input positively impacts the communication development in individuals with complex communication needs.⁴⁷ Future studies could systematically examine the change over time in language development as influenced by the type, frequency, and duration of AAC interventions.

Acknowledgments

We thank the children who participated in this research and their parents who completed the parent-report measures, as well as the graduate and

undergraduate students at the University of Wisconsin – Madison who assisted with data collection and reduction.

Funding

This study was funded by grant [R01DC009411] awarded to Katherine C. Hustad 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 Centre, [U54 HD090256], from the National Institute of Child Health and Human Development, National Institutes of Health. The content is solely the responsibility of the authors and does not necessarily represent the official views of the NIH.

Declaration of Interest

The authors have no financial relationships relevant to this article to disclose. The authors have no conflicts of interest to disclose.

References

1. Vos RC, Dallmeijer AJ, Verhoef M, Van Schie PE, Voorman JM, Wiegerink DJ, Geytenbeek JJM, Roebroek ME, Becher JG. Developmental trajectories of receptive and expressive communication in children and young adults with cerebral palsy. *Dev Med Child Neurol.* 2014;56(10):951–59. doi:10.1111/dmcn.12473.
2. Bax M, Tydeman C, Clinical FO. MRI correlates of cerebral palsy: the European cerebral palsy study. *J Am Med Assoc.* 2006;296(13):1602–08. doi:10.1001/jama.296.13.1602.
3. Hustad KC, Allison K, McFadd E, Riehle KJDN. Speech and language development in 2-year-old children with cerebral palsy. *Int J Lang Commun Disord.* 2014;17:167–75.
4. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol.* 1997;39:214–23.
5. Eliasson AC, Krumlinde-Sundholm L, Rosblad B, Beckung E, Arner M, Ohrvall AM, Rosenbaum P. 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(7):549–54. doi:10.1017/S0012162206001162.
6. Sellers D, Mandy A, Pennington L, Hankins M, Morris C. Development and reliability of a system to classify the eating and drinking ability of people with cerebral palsy. *Dev Med Child Neurol.* 2014;56(3):245–51. doi:10.1111/dmcn.12352.
7. Hidecker MJC, Paneth N, Rosenbaum PL, Kent RD, Lillie J, Eulenberg JB, Chester K, Johnson B, Michalsen L, Evatt M, et al. Developing and validating the communication function classification system for individuals with cerebral palsy. *Dev Med Child Neurol.* 2011;53(8):704–10. doi:10.1111/j.1469-8749.2011.03996.x.
8. Himmelman K, Lindh K, Hidecker MJ. Communication ability in cerebral palsy: a study from the CP register of western Sweden. *Eur J Paediatr Neurol.* 2013;17(6):568–74. doi:10.1016/j.ejpn.2013.04.005.
9. 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(6):1496–513. doi:10.1044/1092-4388(2010/09-0176).
10. Barty E, Caynes K, Johnston LM. Development and reliability of the functional communication classification system for children with cerebral palsy. *Dev Med Child Neurol.* 2016;58(10):1036–41. doi:10.1111/dmcn.13124.
11. Hustad KC, Allison KM, Sakash A, McFadd E, Broman AT, Rathouz PJDM. Longitudinal development of communication in children with cerebral palsy between 24 and 53 months: predicting speech outcomes. *Dev Neurorehabil.* 2017;20(6):323–30. doi:10.1080/17518423.2016.1239135.
12. Hustad KC, Sakash A, Broman AT, Rathouz PJ. Longitudinal growth of receptive language in children with cerebral palsy

- between 18 months and 54 months of age. *Dev Med Child Neurol*. 2018;60:1156–64. doi:10.1111/dmcn.2018.60.issue-11.
13. 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*. 2016;58(6):605–11. doi:10.1111/dmcn.12957.
 14. Pecyna PM, Sommers RK. Testing the receptive language skills of severely handicapped preschool children. *Lang Speech Hear Serv Schools*. 1985;16(1):41–52. doi:10.1044/0161-1461.1601.41.
 15. Geytenbeek J, Harlaar L, Stam M, Ket H, Becher JG, Oostrom K, Vermeulen J. Utility of language comprehension tests for unintelligible or non-speaking children with cerebral palsy: a systematic review. *Dev Med Child Neurol*. 2010;52(12):e267–e77. doi:10.1111/j.1469-8749.2010.03807.x.
 16. Coleman A, Weir KA, Ware RS, Boyd RN. Relationship between communication skills and gross motor function in preschool-aged children with cerebral palsy. *Arch Phys Med Rehabil*. 2013;94(11):2210–17. doi:10.1016/j.apmr.2013.03.025.
 17. Griffiths T, Addison A. Access to communication technology for children with cerebral palsy. *Paediatr Child Health*. 2017;27(10):470–75. doi:10.1016/j.paed.2017.06.005.
 18. Binger C, Maguire-Marshall M, Kent-Walsh J. Using aided AAC models, recasts, and contrastive targets to teach grammatical morphemes to children who use AAC. *J Speech Lang Hear Res*. 2011;54(1):160–76. doi:10.1044/1092-4388(2010/09-0163).
 19. Geytenbeek JJ, Heim MM, Vermeulen RJ, Oostrom KJ. Assessing comprehension of spoken language in nonspeaking children with cerebral palsy: application of a newly developed computer-based instrument. *Augment Altern Commun*. 2010;26(2):97–107. doi:10.3109/07434618.2010.482445.
 20. Geytenbeek JJ, Heim MJ, Knol DL, Vermeulen RJ, Oostrom KJ. Spoken language comprehension of phrases, simple and compound-active sentences in non-speaking children with severe cerebral palsy. *Int J Lang Commun Disord*. 2015;50(4):499–515. doi:10.1111/1460-6984.12151.
 21. Fenson L, Resznick S, Thal D, Bates E, Hartung J, Reilly J. The MacArthur communicative development inventories. San Antonio (TX): Psychological Corporation; 1992.
 22. Bornstein MH, Haynes OM. Vocabulary competence in early childhood: measurement, latent construct, and predictive validity. *Child Dev*. 1998;69:654–71.
 23. Heilmann J, Weismer SE, Evans J, Hollar C. Utility of the MacArthur–bates communicative development inventory in identifying language abilities of late-talking and typically developing toddlers. *Am J Speech Lang Pathol*. 2005;14:40–51.
 24. Dale PS, Bates E, Reznick JS, Morisset C. The validity of a parent report instrument of child language at twenty months. *J Child Lang*. 1989;16:239–49.
 25. Thal DJ, O'Hanlon L, Clemmons M, Fralin L. Validity of a parent report measure of vocabulary and syntax for preschool children with language impairment. *J Speech Lang Hear Res*. 1999;42(2):482–96. doi:10.1044/jslhr.4202.482.
 26. Charman T, Drew A, Baird C, Baird G. Measuring early language development in preschool children with autism spectrum disorder using the MacArthur communicative development inventory (Infant Form). *J Child Lang*. 2003;30:213–36.
 27. Venker CE, Haebig E, Edwards J, Saffran JR, Weismer SE. Brief report: early lexical comprehension in young children with ASD: comparing eye-gaze methodology and parent report. *J Autism Dev Disord*. 2016;46(6):2260–66. doi:10.1007/s10803-016-2747-z.
 28. Miller JF, Sedey AL, Miolo G. Validity of parent report measures of vocabulary development for children with Down syndrome. *J Speech Lang Hear Res*. 1995;38(5):1037–44. doi:10.1044/jslhr.3805.1037.
 29. Vandereet J, Maes B, Lembrechts D, Zink I. Predicting expressive vocabulary acquisition in children with intellectual disabilities: A 2-year longitudinal study. *J Speech Lang Hear Res*. 2010;53(6):1673–86. doi:10.1044/1092-4388(2010/09-0187).
 30. Cuda D, Murri A, Guerzoni L, Fabrizi E, Mariani V. Pre-school children have better spoken language when early implanted. *Int J Pediatr Otorhinolaryngol*. 2014;78(8):1327–31. doi:10.1016/j.ijporl.2014.05.021.
 31. Tomblin JB, Barker BA, Spencer LJ, Zhang X, Gantz BJ. The effect of age at cochlear implant initial stimulation on expressive language growth in infants and toddlers. *J Speech Lang Hear Res*. 2005;48(4):853–67.
 32. 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(6):323–30. doi:10.1080/17518423.2016.1239135.
 33. Dale PS. The validity of a parent report measure of vocabulary and syntax at 24 months. *J Speech Lang Hear Res*. 1991;34(3):565–71. doi:10.1044/jslhr.3403.565.
 34. Smith V, Mirenda P, Zaidman-Zait A. Predictors of expressive vocabulary growth in children with autism. *J Speech Lang Hear Res*. 2007;50(1):149–60. doi:10.1044/1092-4388(2007/013).
 35. Yoder P, Watson LR, Lambert W. Value-added predictors of expressive and receptive language growth in initially nonverbal preschoolers with autism spectrum disorders. *J Autism Dev Disord*. 2015;45(5):1254–70. doi:10.1007/s10803-014-2286-4.
 36. Sabbadini M, Bonanni R, Carlesimo G, Caltagirone C. Neuropsychological assessment of patients with severe neuromotor and verbal disabilities. *J Intellect Disabil Res*. 2001;45:169–79.
 37. 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(3):252–66. doi:10.3109/07434618.2014.924992.
 38. Geytenbeek JJ, Heim MM, Vermeulen RJ, Oostrom KJ. Assessing comprehension of spoken language in nonspeaking children with cerebral palsy: application of a newly developed computer-based instrument. *Augment Altern Commun*. 2010;26(2):97–107. doi:10.3109/07434618.2010.482445.
 39. Benedict H. Early lexical development: comprehension and production. *J Child Lang*. 1979;6:183–200.
 40. Sevcik RA. Comprehension: an overlooked component in augmented language development. *Disabil Rehabil*. 2006;28(3):159–67. doi:10.1080/09638280500077804.
 41. Frank MC, Braginsky M, Yurovsky D, Marchman VA. Wordbank: an open repository for developmental vocabulary data. *J Child Lang*. 2017;44(3):677–94. doi:10.1017/S0305000916000209.
 42. Rowe ML, Raudenbush SW, Goldin-Meadow S. The pace of vocabulary growth helps predict later vocabulary skill. *Child Dev*. 2012;83(2):508–25. doi:10.1111/j.1467-8624.2011.01710.x.
 43. Romski MA, Sevcik RA, Cheslock M, Barton A. The system for augmenting language: AAC and emerging language intervention. In: McCauley F, Paul H, editors. *Treatment of language disorders in children: conventional and controversial intervention*. Baltimore (MD): Paul H. Brookes; 2006.
 44. Biggs EE, Carter EW, Gilson CB. Systematic review of interventions involving aided AAC modeling for children with complex communication needs. *Am J Intellect Dev Disabil*. 2018;123(5):443–73. doi:10.1352/1944-7558-123.5.443.
 45. Dada S, Alant E. The effect of aided language stimulation on vocabulary acquisition in children with little or no functional speech. *Am J Speech Lang Pathol*. 2009;18(1):50–64. doi:10.1044/1058-0360(2008/07-0018).
 46. Harris MD, Reichle J. The impact of aided language stimulation on symbol comprehension and production in children with moderate cognitive disabilities. *Am J Speech Lang Pathol*. 2004;13(2):155–67. doi:10.1044/1058-0360(2004/016).
 47. O'Neill T, Light J, Pope L. Effects of interventions that include aided augmentative and alternative communication input on the communication of individuals with complex communication needs: A meta-analysis. *J Speech Lang Hear Res*. 2018;61(7):1743–65. doi:10.1044/2018_JSLHR-L-17-0132.