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Speech and language development in 2-year-old children with cerebral palsy

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Abstract
Objective: We examined early speech and language development in children who had cerebral palsy. Questions addressed whether children could be classified into early profile groups on the basis of speech and language skills and whether there were differences on selected speech and language measures among groups.
Methods: Speech and language assessments were completed on 27 children with CP who were between the ages of 24 and 30 months (mean age 27.1 months; SD 1.8). We examined several measures of expressive and receptive language, along with speech intelligibility.
Results: Two-step cluster analysis was used to identify homogeneous groups of children based on their performance on the seven dependent variables characterizing speech and language performance. Three groups of children identified were those not yet talking (44% of the sample); those whose talking abilities appeared to be emerging (41% of the sample); and those who were established talkers (15% of the sample). Group differences were evident on all variables except receptive language skills.
Conclusion: 85% of 2-year-old children with CP in this study had clinical speech and/or language delays relative to age expectations. Findings suggest that children with CP should receive speech and language assessment and treatment at or before 2 years of age.

Keywords: *Cerebral palsy, early intervention, speech development, language development*

Introduction

Children with cerebral palsy are at risk for significant speech, language, and communication problems. Such problems can arise from deficits in speech-motor control, cognition, language, sensation/perception, or a combination of these. Estimates have suggested that approximately 60% of school-aged children with CP have some type of communication challenge as determined by physician observation [1]. In our own work, based on detailed speech and language assessment data from a cohort of 4.5-year-old children with CP, 75% of participants had clinical speech and/or language impairments [2]. Communication challenges of any kind can lead to educational and social isolation [3], and can have a detrimental impact on nearly all aspects of development [4–6]. Thus, identifying and treating specific speech and language problems at the earliest possible age is of the utmost importance.

To date, research on speech and language development in children with CP has been limited, in part due to the extreme heterogeneity of this population. The range of possible speech, language, and communication problems is considerable. To reduce this heterogeneity, we developed a rubric for considering different speech and language impairment profiles in children with CP [2]. Our model separates children into profile groups based on the presence or absence of speech motor involvement, the severity of speech motor involvement, and the presence or absence of language/cognitive involvement. The resultant model comprises 8 categorical speech and language impairment profiles. Preliminary work has validated this model on children with CP at the age of 4.5 years [2]. One key challenge with this model is that it is difficult to apply to very young children (below the age of three years) because of the wide range of variability in speech and language performance that

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Table I. Demographics of children with CP.

Child	CA	Adjusted age	Sex	Type of CP	Anatomical involvement	GMFCS	Speech/language cluster
1	28.4	26.0	M	Spastic	Hemiplegia (right)	3	2
2	27.5	NA	F	Spastic	Hemiparesis (right)	1	3
3	26.9	NA	F	Spastic	Not identified	1	2
4	26.6	NA	M	Spastic	Hemiparesis (left)	1	1
5	28.0	NA	M	Spastic	Not identified	4	1
6	24.5	22.1	F	Spastic	Quadriplegia	2	2
7	28.8	NA	F	Spastic	Not identified	5	1
8	29.3	NA	M	Spastic	Quadriplegia	5	1
9	29.9	NA	M	Spastic	Quadriplegia	5	1
10	25.5	NA	F	Spastic	Quadriplegia	5	1
11	26.8	22.9	M	Not identified	Not identified	1	2
12	26.8	22.9	M	Spastic	Quadriplegia	4	1
13	29.4	26.5	M	Spastic	Quadriplegia	5	2
14	26.5	NA	M	Not identified	Not identified	2	2
15	27.6	23.6	F	Spastic	Diplegia	2	2
16	26.9	NA	M	Spastic	Hemiparesis (right)	1	2
17	29.6	27.8	F	Spastic	Diplegia	4	2
18	29.6	27.0	M	Spastic	Quadriplegia	4	3
19	25.0	NA	M	Spastic	Hemiparesis	1	3
20	24.3	NA	F	Spastic	Quadriplegia	5	1
21	27.6	NA	F	Spastic	Quadriplegia	3	2
22	27.6	26.4	M	Spastic, ataxic	Hemiplegia	5	1
23	24.5	22.4	F	Spastic	Diplegia	3	3
24	24.8	NA	F	Spastic, ataxic	Quadriplegia	5	1
25	28.7	25.3	F	Spastic	Quadriplegia	4	1
26	26.5	NA	M	Spastic	Quadriplegia	5	1
27	24.6	NA	F	Spastic	Hemiparesis (right)	2	2

is considered typical in young children. Such variability during the toddler years can make it difficult to definitively identify problems in young children, especially in cases where deficits are more subtle. Over time, however, the range of acceptable variability narrows, making determination of delays or disorders less complex in many cases.

In the present study, we sought to characterize early speech and language abilities in a cohort of young children with cerebral palsy. Given that we know the majority of children with CP show evidence of speech and language impairments later in the preschool years, we wondered whether we could identify those problems earlier so that we could begin to work toward delivering earlier intervention or even prevention of later problems. Because of the expected range of variability among children and the fact that the presence or absence of speech motor involvement (a fundamental differentiator in our classification system for older children with CP) may not yet be discernible at 2 years of age, we used a broader descriptive approach to characterization of early communication abilities. Our specific questions were as follows: (1) What are the speech and language profiles of young children with cerebral palsy? (2) Do children in different profile groups vary with regard to a select set of speech and language measures?

Method

Participants

Twenty-seven children with CP participated in this study. All children were participating in a larger prospective longitudinal study on communication development in children with CP. Children were recruited through local and regional neurology and physiatry clinics. We sought to recruit a representative sample of children with CP that was not biased for or against the presence of speech or language problems. Inclusion criteria for the larger study required that children (1) have a medical diagnosis of CP and (2) have hearing abilities within normal limits as documented by either formal audiological evaluation or distortion product otoacoustic emission screening. For the present study, an additional inclusion criteria required that the children (3) had completed a data collection session within an age interval of 24–29.5 months.

The mean age across the children included in this study was 27.1 months (SD 1.8). The sample comprised 14 boys (mean age 27.7 months (SD 1.5)) and 13 girls (mean age 26.5 months (SD 1.9)). Of these, five girls and six boys were born prematurely (defined as date of birth three or more weeks prior to expected due date). Table 1 presents demographic characteristics of the children, including CP diagnosis, adjusted age (corrected for prematurity based on expected due date), and Gross Motor Function Classification System (GMFCS) [7] rating.

Materials and procedures

The research protocol involved administration of a standard assessment battery focused on speech production, language comprehension, and spontaneous communication. The play-based data collection sessions lasted approximately 90 min; all children tolerated this without difficulty. The protocol was administered by a certified speech-language pathologist in a sound-attenuating room. The same testing room, stimulus materials, and assessment protocol were employed for each child. Parents were invited to be in the room with their child. All sessions were audio and video recorded with professional-quality recording equipment. In addition, prior to each data collection session, parents were mailed a series of questionnaires to complete and return at the time of the session.

In this study, we selected seven different measures that reflect early speech and language development in children, as well as variables that may reflect the unique deficits children with CP face. Five of the variables were obtained through Systematic Analysis of Language Transcripts (SALT) [8] from parent-child interaction. These were: mean length of utterance in morphemes (MLU-M), number of different words (NDW), number of total words (NTW), percent intelligible utterances, and number of vocal utterances. We also examined the number of words produced as indicated by parent report on the Communication Development Inventory (CDI) [9]. Finally, we examined language comprehension scores as obtained from the Preschool Language Scale – 4 (PLS-4) [10]. Details regarding each measure are provided below.

Measures obtained through SALT. Parent–child interaction samples were obtained as part of the data collection protocol. A standard set of toys and books appropriate for children between the ages of 0–36 months was provided in the testing suite for use during the interaction. Parents were instructed to play with their child as they naturally would at home.

Language transcripts of parent-child interaction samples were created using SALT [8]. Samples were transcribed using standard SALT conventions for utterance segmentation, morphology, and unintelligible words/utterances. Because the children had extremely varied communication abilities, and many children were not capable of producing the standard 50 utterance analysis set, we controlled for duration of the interaction, examining 10 min interaction samples for each child.

From the 10 min transcribed interaction samples, the following expressive language variables were computed based on an analysis set of all complete and intelligible utterances from the full transcripts: MLU-M, NDW and NTW. In addition, two measures were obtained using an analysis set that comprised the entire transcript. First, number of vocal utterances (defined as the number of audible utterances, including those that were completely or partially unintelligible) was determined based on the entire transcript. Note that all child vocalizations were counted as vocal utterances for this measure. Situations in which children produced babbling or jargon were not differentiated from vocalizations that appeared to be unintelligible words or word approximations. For this variable, any string of uninterrupted vocalization was coded as a single unintelligible utterance. Although this measure does not differentiate between intentional linguistic communicative attempts and pre-intentional vocal behavior or vocal play, we felt that it captured an important variable, use of vocalizations in the context of a social interaction, which may have longitudinal importance with regard to the development of talking in children with CP.

We also examined the percent of intelligible utterances as a dependent variable of interest. This was defined as the number of complete and intelligible utterances divided by the total number of vocal utterances (and multiplied by 100), based on the entire transcript. This measure was used as a gross index of intelligibility at the utterance level, serving as a substitute for more formal intelligibility assessment [11], which could not be obtained from children at this age due to developmental limitations and task demands.

To ensure that transcription-based data were reliable, interaction samples were randomly selected from 10 different children and were independently transcribed by a second trained transcriber. SALT analysis data on the variables of interest from the first transcription on each child were compared with SALT analysis data from the second transcription for each child. Reliability was determined by calculating the number of agreements over the total number of judgments for each variable of interest across children. Agreement was as follows: MLU-M = 96%, NDW = 95%; NTW = 90%, number of vocal utterances = 96%, and percent intelligible utterances = 95%.

Measures obtained through CDI. The MacArthur CDI was completed by parents of all participants prior to the assessment session. For some families the data collection session between 24 and 29 months of age was their first longitudinal visit, while for others it was the second or third visit. Parents were asked to complete the Words and Gestures form of the CDI at the time of their child's first visit, regardless of age, as we were interested in capturing information about gestures used by children with CP. For each child's second and subsequent visits, the more advanced Words and Sentences form of the CDI was completed by parents. Seventeen of the children in this study contributed CDI data via the Words and Gestures form. Note that the form contains 396 items and none of the children reached a ceiling (highest score was 207). Twelve of the children in this study contributed CDI data via the Words and Sentences form. Note that this form contains 680 items; none of the children reached a ceiling (the highest score was 361, which is lower than the ceiling for the developmentally earlier Words and Gestures form of the instrument). For the present study, expressive vocabulary was measured by the number of words produced as reported on the CDI, regardless of form administered. One parent did not complete a CDI form.

Language comprehension. The receptive language portion of the PLS-4 [10] was administered to characterize language comprehension. The PLS-4 is normed on children between the ages of 2 days and 6 years-11 months and therefore is sensitive to very early skills. Because several of the children in this study had significant motor impairments, standard administration procedures for the PLS-4 were adapted to enable participation in testing for items involving manual manipulation on a child-by-child and item-by-item basis. Instructions in the technical manual were followed for setting up adaptations and consistent adaptations were employed across the children who needed them.

We used age equivalent scores as the dependent measure for language comprehension because they are based on raw scores, are independent of chronological age, and are readily interpretable. For the purposes of this study, age equivalent scores generally reflected a greater range of abilities across children than standard scores. For example, two children who had standard scores of 50 may differ by as much as 6 months in their age equivalent scores. We were interested in preserving these fine-grained differences between children.

Design and analysis

This study used two-step cluster analysis to identify homogeneous groups of children based on their

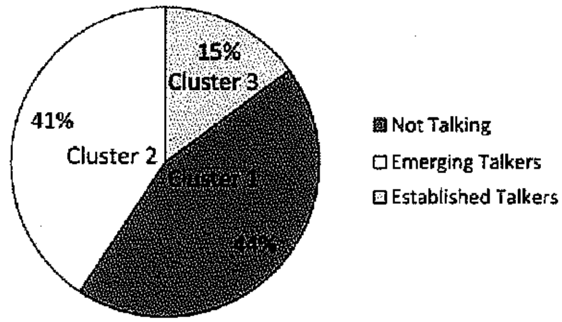

Figure 1 Summary cluster analysis results.

performance on the seven dependent variables. The two-step method identifies pre-clusters in the first step of the analysis and hierarchical clustering in the second step. In this analysis, the number of groups (3) was pre-specified and all variables were entered as continuous. Group membership was then determined through a statistical algorithm that minimized within group variation and maximized between group variation on each of the dependent variables. Pairwise differences between clusters of children on each of the dependent measures were then examined to externally validate the cluster solution. Nonparametric analyses using the Kruskal–Wallis test were performed to examine whether there were differences among the three clusters on each dependent measure. For this analysis, each dependent measure was allotted an alpha level of 0.01; therefore, probability levels less than or equal to 0.01 were necessary for a result to be considered significant. Mann–Whitney U pairwise follow-up tests were then performed for each significant Kruskal–Wallis test to examine differences between the three clusters on each dependent variable. An alpha level of 0.05 was assigned to each family of tests and was partitioned evenly among the three tests. To be considered significant, a probability less than or equal to 0.0167 was necessary for each follow-up test.

Results

Descriptive results for the two-step cluster analysis are shown in Figure 1. Based on the seven different dependent variables, Cluster 1 had 12 children (44% of the sample), Cluster 2 had 11 children (41% of the sample), and Cluster 3 had four children (15% of the sample). Cluster quality was rated 0.6 (on a scale ranging from −1.0 to +1.0), which is considered "good." In order of importance for group differentiation, the top three predictors were: MLU-M, CDI words produced, NDW. Figure 2 illustrates the

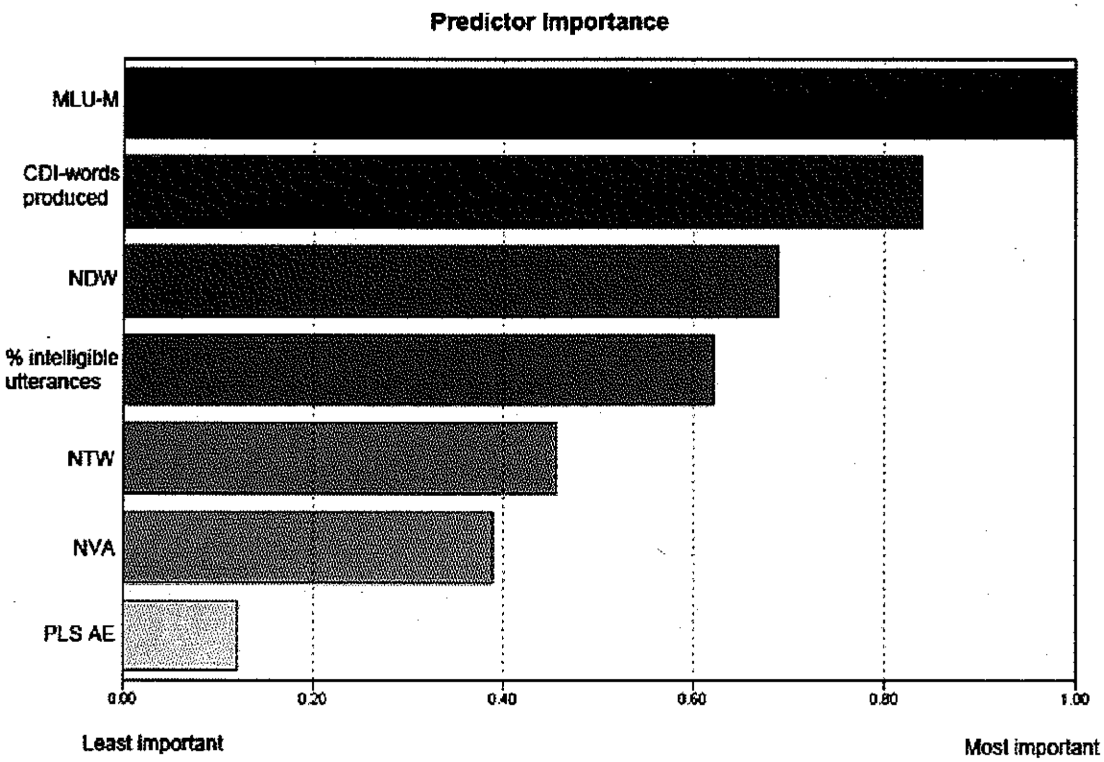

Figure 2 Two-step cluster analysis predictor importance for differentiation into groups.

Table II. Summary results for each dependent variable by Cluster.

	Cluster 1 Not talking $n=12$; 44% Mean (SD)	Cluster 2 Emerging talkers $n=11$; 41% Mean (SD)	Cluster 3 Established talkers $n=4$; 15% Mean (SD)	KW statistic
MLU-M	0	1.14 (0.16)	1.57 (0.45)	23.16*
CDI words produced	2.75 (4.05)	39.54 (39.84)	273.25 (64.37)	18.55*
NDW	0	8.09 (4.95)	37.75 (14.24)	24.02*
% intell utts	0	35.18 (19.44)	70.00 (11.00)	23.63*
NTW	0	20.09 (13.77)	88.50 (53.03)	23.77*
Number vocal utts	21.92 (16.77)	51.09 (16.29)	83.50 (22.09)	15.00*
PLS AE	10.92 (8.35)	19.09 (8.63)	22.00 (1.41)	8.49

Notes: CDI words produced = number of words produced based on parent report via the Macarthur-Bates CDI, % intell utts = percent of intelligible utterances; number of vocal utts = number of vocal utterances produced, regardless of whether the utterances were intelligible; and PLS AE = age equivalent score on the PLS-4. *$p \leq 0.01$.

relative importance of each predictor to cluster membership.

To interpret and name clusters, we examined descriptive data on each dependent variable, shown in Table 2. Data from Cluster 1 clearly suggest that these children had significant limitations in their communication abilities. Children in Cluster 1 had the lowest scores on all of the measures, and had scores of 0 for MLU-M, NDW, NTW, and percent intelligible utterances. Children in this group had an average of 2.75 words per the CDI, and children did produce vocalizations, either in the form of vocal play or unintelligible approximations. Finally, receptive language abilities were variable, with 1 child showing language abilities within normal limits, three showing language abilities between 1 and 2 standard deviations from age expectations, and eight showing language abilities below 2 standard deviations from age expectations. Based on these data, we labeled this group as "not talking."

Data from Cluster 2 suggest that these children had emerging communication skills.

Table III. Mann–Whitney U contrasts examining differences between groups on speech and language variables.

Variable	Pairwise contrast: Not talking *vs.* established talkers	Not talking *vs.* emerging talkers	Established talkers *vs.* emerging talkers
MLU-M	$z = -3.82$*	$z = -4.39$*	$z = -2.38$
CDI	$z = -2.99$*	$z = -3.44$*	$z = -2.87$*
NDW	$z = -3.82$*	$z = -4.39$*	$z = -2.88$*
% intell utts	$z = -3.82$*	$z = -4.38$*	$z = -2.68$*
NTW	$z = -3.82$*	$z = -4.39$*	$z = -2.75$*
Vocal utts	$z = -2.79$*	$z = -3.08$*	$z = -2.22$

Notes: An alpha level of 0.05 was assigned to each family of tests and was partitioned evenly among the three tests. To be considered significant, probability levels less than or equal to 0.0167 were necessary for each follow-up test.
*$p \leq 0.0167$.

Although children in Cluster 2 had relatively few total words and even fewer different words observed in the parent–child interaction sample, they were beginning to combine morphemes, as indicated by an MLU of 1.14, and had an average of approximately 40 words per the CDI. The percent of intelligible utterances was relatively low (35%), however, children produced a larger number of vocalizations, either in the form of vocal play or unintelligible approximations. Again, receptive language was variable, with four children showing language abilities within normal limits, three showing language abilities between 1 and 2 standard deviations from age expectations, and four showing language abilities below 2 standard deviations from age expectations. Based on these data, we labeled this the "emerging talker" group.

Finally, data from Cluster 3 suggest that these children have nicely established communication abilities. Children in Cluster 3 had descriptively higher scores on all measures of speech and language examined in this study, with profiles that were more consistent with typical age-level expectations than the other two groups. Notably, only one of these children had language comprehension scores that did not fall within normal limits, and that child's score fell between 1 and 2 standard deviations of age expectation. Based on these data, we named this the "established talker" group.

For each variable, differences among groups were examined to determine significance. Results revealed that group differences were significant for MLU-M (TS = 23.16; $p < 0.001$)), CDI words produced (TS = 18.55; $p < 0.001$), NDW (TS = 24.02; $p < 0.001$), percent intelligible utterances (TS = 23.63; $p < 0.001$), NTW (TS = 23.77; $p < 0.001$), number of vocal utterances (TS = 15.00; $p = 0.001$). Group differences for language comprehension scores were not significant. Pairwise differences between groups for each of the significant dependent variables revealed that all pairwise differences were significant except for the difference between established and emerging talkers for MLU-M and number of vocal utterances. Statistical results for pairwise contrasts are shown in Table 3.

Discussion

In the present study, we sought to characterize speech and language abilities of 2-year children who had been diagnosed with CP. Results of this study revealed two key findings that will be discussed. First, there were three clear speech and language profile groups among 2-year-old children with CP. Two of those groups comprise children who had clear and significant speech and language impairments and constituted 85% of the sample. The second key finding was that children in the three different profile groups generally showed consistent differences on expressive language and speech variables, but not on receptive language abilities, which were quite varied across groups. Implications of these findings are discussed below.

Speech and language profiles in 3-year-old children

The present study showed that the children with CP in our sample could be divided into three groups, based on speech and language skills at 2 years of age: one group who was not talking (44% of the sample), one group whose talking was emerging (41% of the sample), and one group who comprised established talkers (15% of the sample).

It is noteworthy and concerning that only a very small proportion of children in this study were established talkers who appeared to be developing speech and language skills that were roughly commensurate with age expectations. Conversely, 85% of the children in this study showed clear evidence of a clinical speech and/or language delay at 2 years of age. Our previous work on a similar group of children indicates that by 4.5 years of age, 75% of children have evidence of a clinical speech and/or language impairment. Taken together with our previous work, it appears that approximately 10% of children with CP may outgrow speech/language problems later in the preschool years. However, it is unclear whether these children catch up on their own, or whether intervention leads to advances in skill development that bring children in line with developmental expectations. Descriptive examination of information regarding intervention provided by parents of children in the present study indicated that 18 of 27 participants (66%) were currently receiving speech and language services through birth-3 programs. Further descriptive analysis of these data by profile group revealed that 10 of 12 children (83%) who were not talking, six of 11 children (55%) who were emerging talkers, and two of four children (50%) who were established talkers were receiving speech and language services. These data suggest that children with CP are underserved with regard to speech and language intervention, particularly those who are emerging talkers.

Profile group differences among speech and language variables

In this study, we examined several different variables that have been widely used in the study of child speech and language development, and comparisons between groups on each of these variables generally showed predictable linear patterns of difference between the three groups on all variables except language comprehension abilities. It is noteworthy that all children were producing vocalizations, even those who were in the not talking group. However, the children who were in the not talking group did not produce any vocal utterances that were intelligible, and these children produced fewer total vocal utterances than children in the other two groups who had more advanced expressive communication skills.

Children who were emerging and established talkers did not differ statistically in the number of vocal utterances that they produced and the mean length of their utterances; however, they did differ significantly on the other expressive variables examined in this study (CDI words produced, NDW, percent intelligible utterances, and NTW). Generally, these findings suggest that although children in the emerging talker group had considerably less advanced communication abilities than children in the established talker group, some of their expressive language skills were beginning to look like those of children in the established talker group.

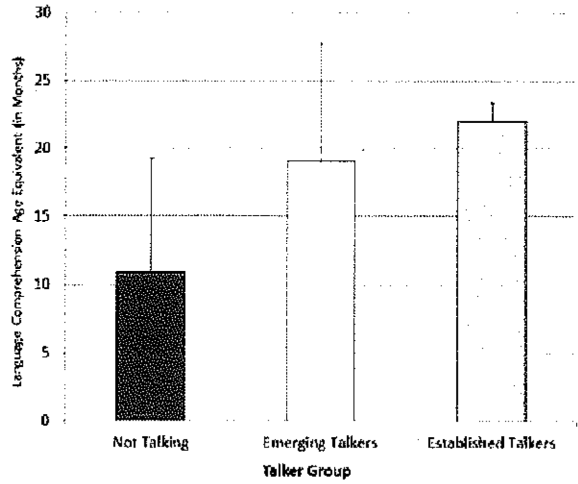

Figure 3 Language comprehension scores by talker group.

Another key finding of the present study was that differences in receptive language ability were not significant across the groups. Figure 3 shows descriptive data regarding language comprehension. One noteworthy feature of this figure is the very large standard deviations within groups, indicating considerable variability among children within and between groups. Examination of individual child data suggests that all children in the established talker group had relatively strong receptive language skills. However, children who were not yet talking and those whose talking was emerging showed a varied composition with regard to language comprehension. This finding is not surprising given that delays in talking can be associated with different origins, including reduced cognitive linguistic skills and/or reduced speech motor abilities.

Definitive connections between speech motor involvement, cognitive/language involvement, and gross motor involvement have not been established. However, several studies have found that children with more severe cognitive involvement may also tend to have more severe gross motor involvement [12, 13]. In an attempt to determine what gross motor function might tell us about speech and language abilities, we examined descriptive data from the GMFCS for each child in this study. Results indicated that several levels of gross motor involvement were represented in each of the three profile groups and that there did not appear to be a consistent pattern of GMFCS scores within the different profile groups (Table 1). This observation suggests that it is important not to draw conclusions regarding language or cognitive abilities on the basis of gross motor involvement.

Limitations

One key limitation of this work is the small sample size ($n=27$). Although, we sought to recruit a representative sample of children with CP, it is possible that our sample may reflect biases of which we are not explicitly aware. For example, parents who had particular concerns regarding their child's communication development may have been more likely to participate in this study than those who did not have concerns. In addition, children who had a diagnosis of cerebral palsy prior to 24 months of age (as all did in the present study) may represent a segment of the population that has more frank deficits than those who are diagnosed later in the preschool years; however, GMFCS data suggest that children in this sample captured a representative range of motor function levels. Additional research is needed to replicate the findings of this study on a larger sample where population-based demographics are available to ensure a representative sampling of participants.

Summary

Children with CP are at significant risk for communication problems. Results of this study indicate that speech and language problems can be identified by 2 years of age in children with CP. Findings suggest that the majority of 2-year-old children with CP may in fact have clinical speech or language delays. Although our data on older children with CP suggest that some children will likely outgrow their communication challenges by time they enter school, most children with CP who have early communication problems will have longstanding and persistent problems with communication as they mature. This finding suggests that treatment should be initiated early for children with CP who show signs of speech or language delay.

Results of this study suggest that all children with CP should receive a comprehensive speech and language assessment by two years of age and those with speech and/or language delays should receive speech and language intervention to enhance the development of speech and language abilities and to enhance their ability to communicate using any and all means available. Specifically, many children with CP and communication challenges would be excellent candidates for interventions [14], including augmentative and alternative communication, to enhance expressive and receptive communication abilities [15].

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The authors alone are responsible for the content and writing of this article.

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