

# NIH Public Access

**Author Manuscript** 

J Speech Lang Hear Res. Author manuscript; available in PMC 2011 December 1

#### Published in final edited form as:

J Speech Lang Hear Res. 2010 December ; 53(6): 1496–1513. doi:10.1044/1092-4388(2010/09-0176).

# Classification of speech and language profiles in 4-year old children with cerebral palsy: A prospective preliminary study

Katherine C. Hustad, Ph.D<sup>\*,+</sup>, Kristin Gorton, M.S.<sup>+</sup>, and Jimin Lee, M.S.<sup>\*,+</sup>

\* Department of Communicative Disorders, University of Wisconsin – Madison

+ Waisman Center, University of Wisconsin - Madison

# Abstract

**Purpose**—Little is known about the speech and language abilities of children with cerebral palsy (CP) and there is currently no system for classifying speech and language profiles. Such a system would have epidemiological value and would have the potential to advance the development of interventions that improve outcomes. In this study, we propose and test a preliminary speech and language classification system by quantifying how well speech and language data differentiate among children classified into different hypothesized profile groups.

**Method**—Speech and language assessment data were collected in a laboratory setting from 34 children with CP (18 males; 16 females) who were a mean age of 54 months (SD 1.8 months). Measures of interest were vowel area, speech rate, language comprehension scores, and speech intelligibility ratings.

**Results**—Canonical discriminant function analysis showed that three functions accounted for 100% of the variance among profile groups, with speech variables accounting for 93% of the variance. Classification agreement varied from 74% to 97% using four different classification paradigms.

**Conclusions**—Results provide preliminary support for the classification of speech and language abilities of children with CP into four initial profile groups. Further research is necessary to validate the full classification system.

## Keywords

cerebral palsy; dysarthria; speech acoustics; speech and language profiles; classification

Cerebral palsy (CP) is the most common cause of severe motor disability in children (Lepage, Noreau, Bernard, & Fougeyrollas, 1998), and it can have a profound impact on all aspects of life (Kennes, Rosenbaum, Hanna, Walter et al., 2002; Liptak, O'Donell, Conaway, Chumlea et al., 2001). For the past 40 years, the prevalence of CP has been relatively stable (or perhaps increasing slightly (Paneth, Hong, & Korzeniewski, 2006)). International estimates suggest that CP affects between 1.2 and 3.0 per 1000 children in developed countries (Odding, Roebroeck, & Stam, 2006; Paneth et al., 2006). However, in the United States, the most recent study suggests that CP may affect up to 3.6 per 1000 children (Yeargin-Allsopp, Braun, Doernbery, Benedict et al., 2008).

CP is an umbrella term for which a number of different definitions have been proposed over the years. The most recent consensus definition specifies that CP: a.) is characterized by movement and posture disturbance; b.) is non-progressive in nature; c.) has its onset within the pre-natal or neonatal period; d.) is caused by some type of damage to the central nervous system; and e.) is often accompanied by co-occurring problems with sensation, perception, cognition, communication, and behavior (Rosenbaum, Paneth, Leviton, Goldstein et al., 2007). Although experts have recognized the latter-most part of this definition for many years, problems with sensation, cognition, communication, and behavior have not been formally acknowledged as core defining aspects of CP until the most recent consensus definition in 2007. Speech and language abilities of children with CP are of particular interest for the present study.

Speech and language problems in children with CP can arise from deficits in speech motor control, cognition, language, sensation/perception, or a combination of these. Recent data from a large population-based sample in Europe indicate that 60% of children with CP have some type of communication problem (Bax, Tydeman, & Flodmark, 2006). The exact nature of these problems has never been systematically studied or classified in children with CP. However, research indicates that communication difficulties can have a variety of adverse affects on children with CP, and can result in differences in social interaction patterns (Light, Collier, & Parnes, 1985a, 1985b, 1985c; Pennington & McConachie, 2001b) and in quality of life (Dickinson, Parkinson, Ravens-Sieberer, Thyen et al., 2007).

Speech deficits, particularly dysarthria, in individuals with CP have received more attention than other communication-related areas. The primary theoretical approach to the study of dysarthria in individuals with CP has emphasized impairment-level differentiation of speech features based on the underlying gross motor movement disorder (Byrne, 1959; Hardy, 1964; Irwin, 1955; Leith & Steer, 1958; Platt, Andrews, Young, & Quinn, 1980; Workinger & Kent, 1991) (i.e. athetoid vs. spastic). Research efforts have focused on identification of acoustic, perceptual, and physiological characteristics of speech, primarily in adults and older children with CP (Byrne, 1959; Hardy, 1964; Irwin, 1955; Leith & Steer, 1958; Platt et al., 1980; Workinger & Kent, 1991). Although some differences in speech features have been identified between individuals with athetoid and spastic CP, there is considerable overlap. Perhaps the most important generalization supported by existing literature is that speech deficits appear to involve all speech subsystems; one important and frequent result is reduced speech intelligibility.

Speech intelligibility has been a topic of considerable interest in the dysarthria literature, with studies examining the relationships between intelligibility and a variety of acoustic and perceptual variables. One well established finding is that there is a strong relationship between articulatory working space (vowel area) and speech intelligibility. Studies of adults, including one examining individuals with CP, have consistently demonstrated that smaller vowel areas are closely related to lower intelligibility ratings (Liu, Tsao, & Kuhl, 2005; Turner, Tjaden, & Weismer, 1995; Weismer, Jeng, Laures, & Kent, 2001). Research has also demonstrated that children with dysarthria of varying etiology tend to have reduced vowel area relative to non-dysarthric speakers (Higgins & Hodge, 2002). However, the relationship between intelligibility and vowel space in children is complicated by the joint influences of speech motor control and vocal tract growth. In particular, studies have demonstrated that as children grow, the vocal tract lengthens and formant frequencies decrease, which ultimately results in decreasing vowel space until about 16 years of age (Vorperian & Kent, 2007). During this same time frame, speech motor control is becoming increasingly refined (Smith & Zelaznik, 2004), which may result in expansion of the vowel space. Meta-analysis of data from typical children suggests that the overall trend of decreasing vowel space with age is comprised of a series of cubic trends (in which vowel space decreases, then increases, then decreases slightly) that span ranges of about 1-3 years, beginning at age 4 (Vorperian & Kent, 2007). Thus, there may be some age specific fluctuation in vowel space as children develop and refine speech motor control. Although

studies of children with speech motor disorders are limited, recent research suggests that children with dysarthria between the ages of 5 and 7 years may have unusually small vowel areas relative to their typically developing same-age peers (Higgins & Hodge, 2002).

CP is a complex disorder that is often accompanied by learning disabilities, problems with cognition, and subsequent reduced language abilities. Recent theories of speech development have emphasized the inter-relation between speech and language development (Smith & Goffman, 2004; Strand, 1992) and speech and cognitive development (Kent, 2004). However, the primary theoretical approach to the study of communication deficits in CP has addressed speech deficits exclusively, with no regard for language or cognition. In fact, individuals with marked language or cognitive deficits have routinely been excluded from studies of speech in CP (see (Achilles, 1955; Ansel & Kent, 1992; Irwin, 1955, 1968; Leith & Steer, 1958; Rutherford, 1939, 1944; Workinger & Kent, 1991) ). Studies suggests that approximately 40% may have reduced cognitive ability, as defined by standardized IQ scores below 70 (Odding et al., 2006; Sigurdardottir, Eriksdottir, Gunnarsdottir, Meintema et al., 2008). Thus, a true account of communication and an inclusive strategy for classifying speech and language abilities of individuals with CP can only emerge from the study of participants who have range of speech, language and cognitive skills.

Extensive research efforts have been directed toward quantification and classification of gross motor (Gorter, Rosenbaum, Hanna, Palisano et al., 2004; Palisano, Hanna, Rosenbaum, Russell et al., 2000; Palisano, Rosenbaum, Walter, Russell et al., 1997b; Rosenbaum, Walter, Hanna, Palisano et al., 2002; Wood & Rosenbaum, 2000) and fine motor (Eliasson, Krumlinde-Sundholm, Rosblad, Beckung et al., 2006; Fedrizzi, Pagliano, Andreucci, & Oleari, 2003; Hanna, Law, Rosenbaum, King et al., 2003; Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006a) development in children with CP. However, the lack of data regarding speech, language and communication development has made it impossible to fully understand the constellation of and inter-relations among deficits. Thus, a comprehensive theory-based understanding of CP with the potential to direct treatment remains elusive. The development and validation of theoretically-driven, research-based tools for classifying speech and language problems in individuals with CP has been identified as a high priority (Bax, Goldstein, Rosenbaum, & Levinton, 2005; Rosenbaum et al., 2007). A speech and language classification system, such as the one we propose in this paper, would have value from an epidemiological perspective in that it would systematize our approach to identification of speech and language deficits and advance our understanding of the specific nature of speech and language disorders observed in children with CP. This would allow for a more comprehensive study of the prevalence of different kinds of speech and language profiles among children with CP. Further, longitudinal study of the stability of classification into profile groups would have important implications for predicting outcomes and potentially for changing or optimizing outcomes via intervention.

In the present study, our aim was to validate a preliminary speech and language classification system for children with CP. We did this by testing how well quantitative speech and language data fit a set of hypothesized speech and language profile groups, and by determining which speech and language variables best differentiated among the hypothesized groups. We also sought to determine how classification into profile groups differed when an exploratory approach was employed relative to when the set of hypothesized groups was employed.

We developed the speech and language classification system based on theoretical foundations regarding the nature of CP and the co-occurrence of deficits (Bax et al., 2005; Bax et al., 2006; Odding et al., 2006; Rosenbaum et al., 2007). We hypothesized that there were eight different communication profile groups among children with CP. See Figure 1.

Because all individuals with CP have a movement disorder, but not all have speech-motor involvement, our first level of differentiation among groups (Level 1) was the presence or absence of speech motor involvement. Speech motor involvement was defined as clinically observable evidence of motor impairment in any one or more of the speech subsystems (articulation, phonation, resonation, respiration) that could be observed perceptually. We hypothesized that at the most superficial level, children with CP could be divided into two groups: those who had speech motor involvement (SMI) and those who had no clinical speech motor involvement (NSMI).

Among the children who had SMI, there was a second level of speech motor differentiation in the model. Within this level (Level 2), we hypothesized that there were two groups: children who were unable to produce speech, as defined by the inability to produce more than 5 differentiable words or word approximations, and children who were able to speak, but who had a speech-motor disorder. Among the children who had a speech-motor disorder but could speak, a further differentiation (Level 2a) based on severity of speech impairment (mild or moderate/severe), was included.

Finally, the model incorporates a third level of differentiation among children with CP based on language and/or cognitive abilities (Level 3). We hypothesized that children could be separated into at least two groups: those with language and/or cognitive abilities within an age appropriate range and those with language and/or cognitive delay relative to age expectations.

From this model, 8 mutually exclusive, categorical profile groups, illustrated in Figure 1, emerged: 1.) children with no clinical speech-motor involvement and typically developing language and/or cognitive abilities (NSMI-LCT); 2.) children with no clinical speech-motor involvement and impaired language and/or cognitive abilities (NSMI-LCI); 3.) children with mild speech-motor impairment and typically developing language and/or cognitive abilities (SMI-MILD-LCT); 4.) children with mild speech-motor involvement and impaired language and/or cognitive abilities (SMI-MILD-LCT); 5.) children with moderate/severe speech-motor involvement and typically developing language and/or cognitive abilities (SMI-MILD-LCI); 5.) children with moderate/severe speech-motor involvement and typically developing language and/or cognitive abilities (SMI-M/S-LCT); 6.) children with moderate/severe speech-motor involvement and impaired language and/or cognitive abilities (ANAR-LCT); and 8.) children with anarthria and impaired language and/or cognitive abilities (ANAR-LCI). Note that within this classification system, speech-motor involvement was regarded as a primary differentiator among groups, and language/cognitive abilities were considered secondary differentiators.

Because we had a relatively small number of participants in the present study, we examined a preliminary, collapsed version of the full classification scheme. Specifically, we classified children into four groups as follows: a.) all children who had no clinical speech-motor involvement were classified together into one group (NSMI); b.) children with speech-motor impairment and age appropriate language and/or cognitive abilities were grouped together regardless of the severity of their speech-motor impairment (SMI-LCT); c.) children with speech-motor impairment and impaired language and/or cognitive abilities were grouped together regardless of the severity of their speech-motor impairment (SMI-LCT); and d.) all children who had anarthria were classified together into one group (ANAR). We collapsed the full classification scheme in this particular way because we felt that it represented the most important distinctions in speech and language ability profiles among the children in our cohort. Figure 2 provides a schematic diagram of the 4 collapsed profile groups. Table 1 provides a description of the groups examined in this study.

The following specific research questions were addressed:

- **1.** Are there differences among children in the four hypothesized communication profile groups on a select set of speech and language measures?
- **2.** If there are differences among groups on the selected measures, which speech and language variables best differentiate the groups and optimize classification of children into the hypothesized groups?
- **3.** When there are no a priori hypothesized groups, how does classification of children into profile groups compare with membership in the hypothesized groups?

## Method

#### **Participants**

Participants in this study were selected from a cohort of children with cerebral palsy who were participating in a 4-year prospective longitudinal study of communication development. In order to examine a sample that was as homogeneous as possible with regard to chronological age, data from the youngest common age across participants, 54 months, were employed for the present study. Criteria for inclusion in 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. To be included in the presented study, an additional criterion required that children: 3.) had completed a data collection session at an average age of 54 months (+/- 4 months). Children with hearing impairment were specifically excluded from this study because of unique issues related to speech production, speech perception, and expressive/receptive language. There were no other exclusion criteria for the study.

Children were recruited through a regional CP clinic, and through local and regional physicians. In addition, children were recruited through birth-to-three service providers and early intervention programs. Because there are no central registries of children with CP in the United States, and because of medical, educational, and research confidentiality restrictions, we were unable to ascertain the total number of families with eligible children who received information regarding the study. Therefore, it is difficult to determine the extent to which our participants represent the population of children with CP. However, given the range of types of CP, functional gross motor skills, and visual impairments among children within our sample (see Table 2), it seems likely that our sample is a representative one. Note, however, that none of the children in our sample had autism spectrum disorders based on both parent report and review of medical records; although in a larger sample it is very likely that children with CP who also have autism spectrum disorders would be represented (Kilincaslan & Mukaddes, 2008).

A total of 34 children (of a possible 40) from the larger study met inclusion criteria. The mean age across the children for the data point used in this study was 54.4 months (SD 1.8). The sample was comprised of 18 boys (mean age 54.6 months (SD 1.6)) and 16 girls (mean age 54.3months (SD 1.9)). Demographic characteristics of children, physiological classification of CP, functional gross motor classification<sup>1</sup> (Palisano, Rosenbaum, Walter, Russell et al., 1997a), and presence of visual impairment by hypothesized communication profile group is provided in Table 2.

<sup>&</sup>lt;sup>1</sup>Physiological classification was made by physicians and was obtained from the children's medical records. Gross motor function classification using the Gross Motor Function Classification System (GFMCS) was obtained by observation during the session and by parent report, both of which have established reliability and validity (Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006b).

J Speech Lang Hear Res. Author manuscript; available in PMC 2011 December 1.

#### **Data Collection Procedures**

The research protocol was administered by a certified speech-language pathologist, the second author, in a sound-attenuating room. The same testing room, stimulus materials, and assessment protocol were employed for each child. In each session, speech production data were collected first, followed by language data. Data collection sessions lasted approximately two hours. Children were offered breaks throughout the sessions, and all children were able to tolerate the sessions without difficulty. 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 order to evaluate our hypothesized set of profile groups, it was necessary to assign children into the mutually exclusive groups. Statistical analysis procedures, described below, were then used to determine whether the differences among groups, per the empirical data, were consistent with our theoretical approach. Following the 54 month assessment session, children were independently classified into one of the four hypothesized communication profile groups by the first two authors based on *clinical impressions* of speech and language performance during the session (and without access to empirical data). The first author observed all sessions, either live or via video-recording; the second author performed all assessment procedures during each session. Decision-making rubric for assignment into groups is shown in Appendix A. Agreement between the first two authors with regard to assignment into groups was 94%; disagreement occurred for 2 of 34 children. Disagreements for both children were focused around whether or not the child had language impairment, with both children having borderline language skills. Disagreements were resolved using discussion and consensus.

#### **Data Reduction: Dependent Measures**

Although many different measures were obtained for each child, we selected four measures to include for quantitative analysis. Measures were chosen on the basis of their clinical relevance and precedence in the research literature. It is important to note that we deliberately limited the variables included in this study to maintain the statistical validity of our analyses in light of the relatively small number of research participants. Toward this end, four different dependent measures representing speech motor abilities and language comprehension were analyzed for each child. Measures reflecting speech motor abilities were: vowel area, speech rate, and speech intelligibility ratings. Standardized language scores were used to reflect language comprehension abilities. Details regarding each measure are provided below.

**Vowel area**—Measures of vowel area, or vowel space, were used to make inferences about articulatory working space. The study of vowels is of value in children with CP because even children with limited speech production ability or reduced speech intelligibility may be able to approximate some vowels (see (Pennington & McConachie, 2001a)). Further, problems with production of vowels have been associated with reduced speech intelligibility in individuals with CP (Ansel & Kent, 1992; Higgins & Hodge, 2002; Liu et al., 2005), and differences in a variety of spectral acoustic variables (e.g. vowel duration, vowel space, F2 slope) have been shown to differ between speakers with and without dysarthria. Thus, we expected that vowel space would play an important role in differentiating among profile groups of children with CP.

Vowel area measures for children who were able to produce speech (Profile Groups NSMI, SMI-LCT, and SMI-LCI) were obtained in the present study by making a series of spectral acoustic measures from digital speech samples using a wideband spectrographic display in

TF 32 (computer software) (Milenkovic, 2002), following established measurement criteria (Kent & Read, 2001; Kent, Weismer, Kent, Vorperian et al., 1999; Klatt, 1976; Turner et al., 1995; Weismer, Laures, Jeng, & Kent, 2000). Children produced a corpus of single word stimuli from the Test of Children's Speech (TOCS) (Hodge & Daniels, 2007), consisting of thirty different words that were lexically and phonetically appropriate for young children. The integrity of productions was monitored by the researcher collecting the data and by a graduate student who was controlling the audio recording equipment. From the corpus of 30 words, eight different stimulus words containing corner vowels were subjected to acoustic analysis. All stimulus words that were used for acoustic analysis were monosyllabic in a consonant-vowel-consonant structure (e.g., target words containing corner vowels: Sheet, Seat; Hoot, Boot; Top, Hot; Bad, Hat). Between two and four repetitions of each of the eight target words were obtained from each child, depending on the child's level of compliance. For each stimulus production, the following measures were made: a) duration of the *phonemes/*i/,/æ/,/a/, and/u/was determined by measuring the interval between the first and last glottal pulse where both F1 and F2 were visible on the spectrogram; b) F1 and F2 frequencies, for the phonemes/1/,/æ/,/a/, and/u/were determined using both wideband spectrographic and spectrum displays from a 30 msec window at the temporal midpoint of each vowel. Spectrogram analysis bandwidth was adjusted for each child based on his/her fundamental frequency (F0) (as determined by the TF32 average F0 algorithm). A bandwidth that was approximately double the child's F0 was employed for analysis. Linear predictive coding was used to generate formant tracks which were hand corrected, as necessary, based on visual inspection of the overlaid tracks on the spectrographic display.

Up to eight tokens (mean = 4.7; SD = 2.9) of each corner vowel were then averaged within each individual child. Computations based on spectral data were made from averages across tokens for individual vowels, and children. Vowel space was calculated using a formula for the area of a polygon in the F1/F2 plane:  $\frac{1}{2}(F1/i/*F2/u/-F1/u/*F2/i/) + \frac{1}{2}(F1/u/*F2/A/-F1/A/*F2/u/) + \frac{1}{2}(F1/A/*F2/u/) + \frac{1}{2}(F1/A/*F2/u/) + \frac{1}{2}(F1/a/*F2/u/) + \frac{1}{2}(F1/a/*F2/u/) + \frac{1}{2}(F1/a/*F2/u/)$  (Johnson, Flemming, & Wright, 2004).

Inter and Intra-judge reliability was obtained for all spectral measures. Intra-judge reliability involved having the same judge make a second set of acoustic measures on all productions from 5 of the 23 (22%) children who were able to speak. Children targeted for reliability were randomly selected from the pool of children who were able to speak, with 1 child selected from Profile Group NSMI, 2 selected from Profile Group SMI-LCT, and 2 selected from Profile Group SMI-LCI. The time lag between the first and second set of measurements was 3 months. The Pearson product-moment coefficient for the first and second set of measurements were 20.16 Hz for F1; and 24.94 Hz for F2. Inter-judge reliability involved having a second judge, who was trained in acoustic analysis methods, evaluate all productions from the same 5 of 23 children targeted for intra-judge reliability. The Pearson product-moment correlation coefficient for the measurements made by the first and second judges was, again, over .99. Absolute differences between measures were 30.06 Hz for F1; and 31.58 Hz for F2. Reliability measures were within an acceptable range following Kent et al. (1999).

**Speech rate**—Individuals with motor speech disorders often have reduced speaking rate (LeDorze, Ouellet, & Ryalls, 1994; Weismer et al., 2000), which is both a characteristic of the disorder and also a compensatory strategy for increasing speech intelligibility (Yorkston, Beukelman, Strand, & Bell, 1999a). Studies of typically developing children have documented that children produce speech more slowly than adults, and that rate of production gets faster with age (Kent & Forner, 1980; Smith, Goffman, & Stark, 1995; Smith, 1978; Smith & Kenney, 1998). Characteristics of speech rate in children with

dysarthria are unknown, but it is likely that they are consistent with findings from the adult dysarthria literature.

In this study, we used overall speaking rate (inclusive of pauses) to provide a general index of speech motor timing and speech subsystem coordination. Reduced speech rate has been associated with both slower articulation rate and with longer pauses between words (Hustad & Sassano, 2002), with both tendencies seeming to become more pronounced as severity of the dysarthria worsens. For this reason and to maintain the ecological validity of the connected speech produced by the children, we chose to include pauses in our measurement of speech rate. Ultimately, we expected that rate would play an important role in differentiating children who had speech-motor involvement from those who did not.

Speech rate was quantified by calculating syllables produced per minute for each of five utterances produced by all children who were able to speak (Profile Groups NSMI, SMI-LCT, and SMI-LCI). Children produced a corpus of utterances from the Test of Children's Speech (TOCS) (Hodge & Daniels, 2007). Utterances ranged in length between 2 and 7 words, and were comprised primarily of mono- and bi-syllabic words. Utterances were obtained by having the child repeat the clinician's model, which was delivered via a recorded set of utterances played on a computer to ensure that all children received the same model. This methodology employing repetition of utterances has been well established in dysarthria research and in our previous work (Hustad, 2007; Hustad & Lee, 2008). Children produced 10 utterances of each length (i.e. ten 2-word utterances, ten 3-word utterances, ten 4-word utterances, and so on) up to the habitual mean length of utterance in words observed in spontaneous speech<sup>2</sup>. The integrity of productions was monitored by the researcher collecting the data and by a graduate student who was controlling the audio recording equipment. Children were asked to repeat any utterances that did not include all target words (or approximations thereof). The first five utterances of the longest length produced by the child were used to calculate speech rate.

Duration of utterances was determined by measuring the time between the onset of audible or visible (on the waveform and spectrogram display) acoustic energy associated with production of the first phoneme of the stimulus utterance and the offset of acoustic energy associated with production of the last phoneme of the stimulus utterance. Speech rate was calculated by dividing the total number of syllables produced by the total duration of production and multiplying this value by 60.

Inter and intra-judge reliability was obtained for temporal measures. Intra-judge reliability involved having the same judge make a second set of acoustic measures on all productions from the same 5 of 23 (22%) children who were targeted for reliability analysis for spectral measures. The time lag between the first and second set of measurements was 3 months. The Pearson product-moment coefficient for the first and second set of measures was over .99. The absolute difference between the first and second measurements was an average of 12 msec (SD 13 msec). Inter-judge reliability involved having a second judge, who was trained in acoustic analysis methods, evaluate all productions from the same 5 of 23 children. The Pearson product-moment coefficient for the measurements made by the first and second raters was, again, over .99. The absolute difference between measures made by the two judges was 14 msec (SD 14 msec). Reliability measures were within an acceptable range following Kent et al. (1999).

 $<sup>^{2}</sup>$ Habitual mean length of utterance was determined informally based on conversational interaction with the child during play with either the clinician or a parent in the data collection session.

J Speech Lang Hear Res. Author manuscript; available in PMC 2011 December 1.

**Speech intelligibility**—Ratings of speech intelligibility were used to provide an indication of the functionality of each child's speech (Yorkston et al., 1999a) and as an index of severity of the speech disorder (Weismer & Martin, 1992). In this study, ratings were obtained from parents who were asked to estimate how intelligible they thought their child's speech was to unfamiliar communication partners. Ratings were made on a 7-point equal appearing scale, where 1 = difficult or impossible to understand, and 7 = very easy to understand. This type of measurement was used, most notably, in the hallmark work of Darley and colleagues, who used equal appearing interval scales to quantify intelligibility of adults with dysarthria (Darley, Aronson, & Brown, 1969).

Parents were used as raters because we were interested in obtaining a measure that reflected the perceptions of the child's most frequent and most familiar communication partner. Parent report is a standard tool in evaluation of child behavior in both clinical practice and in research. The validity of parent report measures has been established in a number of developmental domains including cognition (Johnson, Marlow, Wolke, Davidson et al., 2004) language (Fenson, Resznick, Thal, Bates et al., 1993; Pan, Rowe, Spier, & Tamis-LeMonda, 2004), gross motor function in children with CP (Morris, Galuppi, & Rosenbaum, 2004; Morris et al., 2006b), and feeding and/or oromotor status in children with CP (Gangil, Patwari, Aneja, Ahuja et al., 2001; Reilly, Skuse, & Poblete, 1996). Although parent report of speech intelligibility has received very limited research attention, one study examining parent estimates of their child's intelligibility to *other* people (unfamiliar partners), as we used in the present research, showed findings that were consistent with speech language pathologists' assessment results (Coplan & Gleason, 1988).

**Language comprehension**—Standardized language comprehension scores were used to characterize language and, indirectly, as a gross index of cognitive development. Scores were obtained from one of two standardized tests, depending on developmental capabilities of each child. For all children, the *Test of Auditory Comprehension of Language-Third Edition (TACL-3)* (Carrow-Woolfolk, 1999) was attempted. An important advantage of this test was that completion required very limited motor skills as response options were presented in a field of three discrete pictures which could be selected using manual direct selection or partner directed scanning. In addition, the TACL has three subtests examining receptive vocabulary, understanding of grammatical morphemes, and understanding of elaborated sentences; thus it provides a broader picture of language comprehension beyond simple receptive vocabulary. Twenty-three children completed the TACL.

For children who were unable to understand pictorial representations and were thus unable to attain a basal on the TACL-3, the *Preschool Language Scale-4 (PLS-4)* (Zimmerman, Steiner, & Pond, 2002) was used. The PLS-4 assesses earlier acquired skills and portions of it can be administered via parent interview. On a child-by-child and item-by-item basis, standard administration procedures for the PLS-4 were adapted to enable participation in testing for items involving manual manipulation. Instructions in the technical manual were followed for setting up adaptations and consistent adaptations were employed across the children who needed them. However, for children with significant motor impairment, it was sometimes difficult to discern whether failures were due to lack of understanding of the concept being tested or because the item simply could not be adapted sufficiently to accommodate the child's motor limitations. Thus, for children who were unable to complete the TACL-3 and thus required the PLS-4, language comprehension scores provide only a gross indicator, and likely an underestimate, of language comprehension ability. Eleven children completed the PLS-4.

Because both language measures were standardized tests, reliability and validity of the instruments as established during development of the tests were taken at face value.

Additional reliability data relating to children's performance on standardized language tests, which in some cases involved adaptations to enable accessibility, were not obtained as part of this study.

Scores on the two different language tests were converted to standard scores following the test manuals to permit comparison between the two different tests. Standard scores for both tests were based on a mean of 100 and a standard deviation of 15.

#### **Experimental Design and Analysis**

This study employed a between subjects design to examine differences between the hypothesized communication profile groups on each of the four different dependent measures. Samples sizes for the four profile groups were small, therefore non-parametric analyses using the Mann-Whitney U statistic were performed to evaluate pair-wise differences between groups on each measure. Because this study was exploratory, all comparisons were two-tailed and an alpha level of .01 per contrast was employed.

To determine how well the different speech and language variables separated children into the communication profile groups, and to determine which specific variables were the best predictors of profile group membership, canonical discriminant function analysis was performed. Canonical discriminant function analysis was well suited for our research questions because it does *not* assume that the hypothesized groups are empirically distinct, mutually exclusive, real-world entities. Instead, the hypothesized groups are evaluated on the discriminant functions contain (Cocozzelli, 1988). Thus, this type of analysis enabled us to evaluate whether our hypothesized groups were actually borne out by quantitative data, and to determine which variables (or dimensions comprised of multiple variables) contributed most to group membership. In evaluating the significance of discriminant functions, alpha levels of .05 or less were used. In addition, Box's Test of Equality of Covariance Matrices confirmed that the assumptions of normality and equality of covariance were met.

Finally, cluster analysis was used to identify groups of children without the constraints of the hypothesized profile groups. In this analysis, the number of groups (4) was pre-specified; group membership was then determined through a statistical algorithm that minimized within cluster variance and maximized between cluster variance on each of the four dependent variables. Results of this analysis were intended to be primarily descriptive in nature, and were used as a basis of comparison to validate the pre-specified theoretical model being examined in this study.

# Results

Descriptive results for each of the four dependent variables by communication profile group are shown in Figures 3–6. For each of the four dependent measures, descriptive results followed the predicted pattern according to communication profile group.

Pairwise comparison results examining profile group differences on each of the four dependent measures are shown in Table 3. Results for vowel space showed that all pairwise differences were statistically significant except for the difference between Groups SMI-LCT and SMI-LCI. Results for speech rate followed the same pattern as those for vowel space. Note that for both vowel space and speech rate, comparisons are not reported between Group ANAR and the other groups because valid speech production data were not obtained on these measures. Results for speech intelligibility showed that all comparisons were significant except for the difference between Groups SMI-LCT. Results for speech intelligibility showed that all comparisons were

language comprehension showed that all pairwise differences were significant except for the difference between Groups NSMI and SMI-LCT.

Results of the canonical discriminant function analysis revealed that 100% of the variance was accounted for by three canonical discriminant functions (p<.001). Function 1 accounted for 93.1% of the variance (eigenvalue = 15.19; canonical correlation = .969). Function 2 accounted for an additional 5.6% of the variance (eigenvalue = .91; canonical correlation = . 690). Function 3 accounted for 1.4% of the variance (eigenvalue = .222; canonical correlation = .426). Discriminant functions were interpreted by examining correlations between variables and standardized canonical discriminant functions. Function 1 was comprised of speech rate and vowel space, with speech rate having a higher correlation with the discriminant function (.671) than vowel space (.497). Function 2 was comprised of language comprehension scores. Function 3 was comprised of speech intelligibility ratings. A chart displaying the first two functions and the group centroids for each of the four profile groups is shown in Figure 7.

Overall classification accuracy into the hypothesized groups, based on the canonical discriminant functions, was 97.1% (33 of 34 children). Children from Groups NSMI, SMI-LCI, and ANAR were classified with 100% accuracy (25 of 25 children). Children from Group SMI-LCT were classified with 88.9% accuracy (8 of 9 children).

Cross-validation procedures for classification using a leave-one-out analysis paradigm, in which each case is classified based on all cases *except* the given case, revealed a canonical discriminant function classification accuracy of 91.2% (31 of 34 children) relative to the hypothesized model. Children from Groups SMI-LCI and ANAR were classified with 100% accuracy (17 of 17 children) using the cross-validation procedure. Children from groups NSMI and SMI-LCT were classified with 87.5% accuracy (7 of 8 children) and 77.8% accuracy (7 of 9 children), respectively.

Cluster analysis into four groups with no a priori profile memberships specified resulted in the assignment of 5 children to the first cluster; 10 children to the second cluster; 7 children to the third cluster; and 12 children to the fourth cluster. Point-by-point membership in clusters for each individual child was then compared descriptively with membership in the hypothesized a priori assigned groups. Results indicated that that 79% (27 of 34) of the children were classified into the same group as in our hypothesized model. Relative to our model, classification consistency using cluster analysis was as follows: 62.5% (5 of 8) of the children from our hypothesized Group NSMI were classified into a group together; 67% (6 of 9) of the children from our hypothesized Group SMI-LCT were classified into a group together; and 100% (11 of 11) of the children from our hypothesized Group ANAR were classified into a group together.

Point-by-point agreement for classification of each individual child between each pair of classification methods (a priori assignment into hypothesized groups, canonical discriminant function classification, cross-validated canonical discriminant function classification, and cluster analysis) is shown in Table 4. Data indicate that agreement ranged from a low of 74% (cross-validated canonical discriminant function classification and cluster analysis) to a high of 97% (canonical discriminant function classification and a priori assignment into hypothesized groups). Agreement across all four classification methods was 74%.

## Discussion

In this study, we sought to validate a preliminary speech and language classification system for children with CP. We did this by testing how well quantitative speech and language data

fit the hypothesized speech and language profile groups that comprised our classification system, and by determining which speech and language variables best differentiated among the hypothesized groups. We also sought to examine how classification into profile groups differed when an exploratory approach was employed relative to when the set of hypothesized groups was employed.

#### Differentiating among profile groups

Statistical results provided preliminary support for the presence of the four basic communication profile groups, reflecting a collapsed version of our larger classification system. Results showed that one discriminant function accounted for the vast majority of the variance among children. This function was comprised of speech variables (vowel space and speech rate). Somewhat surprising was that speech rate was more closely correlated with profile group membership than vowel space. One explanation may be tied to the consistent finding of deficits across speech subsystems in individual with CP. Speech rate is a measure that provides an overall index of speech production capability, which inherently involves the integration of all subsystems. Vowel space, however, is more closely tied to articulation, an area where prominent deficits have been documented in individuals with CP. Given the more focused and specific information provided by vowel space data, findings of the present study suggest that it may not adequately reflect the broader range of speech production deficits in children with CP.

Vowel space and speech rate differences among the profile groups comprised of children who were able to speak followed the predicted pattern, with children who had SMI having smaller vowel areas and slower speech rates than children with NSMI. For vowel space, data from children with SMI (Profile Groups SMI-LCT and SMI-LCI) were roughly consistent with previous findings from children who were 5 to 6 years of age with dysarthria, (Higgins & Hodge, 2002). Findings from children with CP in Profile Group NSMI, who did not have evidence of clinical speech motor involvement, were compared with existing literature examining vowel space in typically developing children (see (Higgins & Hodge, 2002; Lee, Potamianos, & Narayanan, 1999) and speech rate (via recitation) in typically developing children (Walker, Archibald, Cherniak, & Fish, 1992). Results suggest that children in group NSMI in the present study had slightly larger vowel spaces than typically developing children in previous studies and slightly slower speech rates. One explanation for the vowel space discrepancy is that previous studies examined children who were older by 6-18 months than those in the present study. Younger children would be expected to have larger vowel spaces because they have smaller vocal tracts and thus have higher formant frequencies (Kent & Read, 2001). Another explanation is that previous studies have employed different speaking tasks and speech stimuli; measurement of both spectral and temporal aspects of speech may have been affected by characteristics of the speech stimuli. Finally, it is possible that some of the children with CP who did not have clinically identified speech motor involvement did, in fact, have subtle differences in their speech production or in the anatomy of their vocal tracts (e.g. higher palatal arches have been anecdotally observed in children born prematurely, as is often the case in children with CP). Further investigation that includes a control group of typically developing children producing the same speech stimuli under the same experimental conditions as employed in the present study is necessary to make accurate comparisons between children with CP and typically developing children.

In addition to speech variables, language comprehension scores also contributed to the differentiation among profile groups, but to a much lesser extent than speech data. This finding supports our hypothesis that speech motor involvement would be a primary differentiator among groups, and language abilities would be as a secondary differentiator. With regard to differences among profile groups on language variables, one surprising

finding was that children in Group ANAR had reduced language abilities relative to their counterparts in Group SMI-LCI. Because the ANAR group was theoretically comprised of children with and without language impairment in the present study, we expected the mean language score across children potentially to be higher than the mean score for children in Group SMI-LCI, all of whom by definition had language impairment. However, examination of individual standard scores for children in Group ANAR indicated that all had clinical language impairment. Potential explanations include the small sample size of the present study and testing limitations for children in Group ANAR, all of whom had severe motor impairment and quadriplegia. Although, there is evidence in the literature to suggest that children with more severe motor involvement also tend to have more severe cognitive involvement (Sigurdardottir et al., 2008), it is universally the case that these children are difficult to assess reliably because of the motor requirements associated with language and cognitive testing. In the present study, test items were adapted as necessary; however, some items could not be adapted sufficiently to accommodate motor challenges, such as access and fatigue, faced by some of the children. In turn, this may have influenced the ability of some children to demonstrate language comprehension skills. Thus, language findings for Group ANAR should be regarded with caution.

Finally, intelligibility ratings made only a very small contribution to differentiation among profile groups. This finding was surprising in light of the importance of speech rate, because intelligibility is also a more global indicator of the integration of speech subsystems (Yorkston, Beukelman, Strand, & Bell, 1999b). One explanation relates to the way that intelligibility was measured in the present study. Use of Likert-scale ratings limited the range and nature of intelligibility data, and parents may have had a biased view of their child's performance. However, findings regarding group differences showed that parent ratings differed among groups in the expected directions for speech intelligibility data. Additional research is necessary to examine the extent to which parent ratings of intelligibility are similar to and different from more traditional scaling and orthographic transcription methodologies employing unfamiliar listeners.

#### **Classification into profile groups**

Results showed that the four variables included in this study resulted in classification into the four speech and language profile groups with accuracy ranging from 74% to 97%, across four classification methods (a priori assignment into groups; canonical discriminant function classification; cross-validated canonical discriminant function classification; and cluster analysis). Thus, collectively, the variables selected to represent speech and language capabilities appeared to be sensitive to group differences among children. It is noteworthy, however, that agreement between cluster analysis (which did not employ a priori groupings) and each of the three other methods (which did make use of the a priori groupings) resulted in the lowest classification agreement values (ranging from 74 to 79%). Discrepancies between methods tended to involve the same children, and the majority were associated with differentiation between children in Group NSMI and Group SMI-LCT. This finding highlights the clinical challenge associated with identifying speech motor disorders in children whose production deficits may be subtle or very mild. The addition of other speechrelated measures such as transcription intelligibility scores, measures reflecting prosody, or hypernasality may aid in the differentiation among children with and without SMI. Classification discrepancies were also noted, but to a lesser extent, between children in Group SMI-LCT and Group SMI-LCI. This observation suggests that additional indices of language and/or cognitive ability may be necessary to aid in the differentiation of children who, again, may have subtle deficits in their language skills.

#### Limitations and future directions

This study is the first of its kind to examine prospective speech and language data from a cohort of children with CP within a very narrow age range. This study is also the first to propose a speech and language classification system for children with CP. Accordingly, findings are preliminary, and conclusions should be drawn with caution for several important reasons, detailed below.

Measurement of language abilities was difficult for children with severe upper extremity involvement. As a result, language skills may be underestimated for some children, particularly those in Group ANAR. In addition, measurement of language comprehension using standardized tests provides only a snapshot into language abilities. Measurement of other aspects of language is necessary to obtain a more comprehensive picture of underlying language and cognitive abilities. Development and evaluation of innovative tools for measuring language in children with CP is necessary. Examples of such tools include eye tracking technology and adaptation of infant preferential gaze paradigms.

Cognitive abilities were not measured in the present study. However, a reasonable assumption is that language comprehension scores roughly approximate cognitive abilities for most children. Similar scores on the TACL and on certain standardized measures of intelligence have been documented in research examining typically developing children and low birth weight children (Kilbride, Thorstad, & Daily, 2004), and children with language delay (Cole & Mills, 1997; Cole, Mills, & Kelley, 1994). Studies employing tools that measure different aspects of cognitive development are necessary to understand the interrelations between speech, language, and cognition among children with CP.

This study examined speech and language profiles of children at the age of four years. Longitudinal studies that examine communication profile group membership using the proposed classification system are necessary to determine whether profile groups are stable over time as children develop, and to determine the earliest age at which speech and language skills can be classified. Such research would have the ability to advance our knowledge regarding the rates and limits of change in speech and language development among children with CP. This information would have critical implications for speech and language interventions.

In this study, classification of children into profile groups was based on clinical observation by two experts, which was then empirically validated using different statistical classification strategies. Agreement between classification strategies varied, suggesting that classification may not be entirely straightforward, particularly for children whose speech and language deficits may be borderline. Additional variables should be considered as contributors to differentiation among groups in future studies.

Ultimately, the utility of the proposed classification system depends directly on whether humans can observe a child and make a reliable classification. Underlying this assumption is that quantitative data would result in the same classification as bestowed by the human observer. The skills of the observer, the duration of the observation, and the particular speech and language tasks that the observer should base his/her classifications upon require study. Further, the study of reliability among different observers is necessary to develop a tool that is both practical and informative.

The relatively small number of participants in this study and the subsequent small number of children comprising each profile group limit generalization of findings. In addition, because there are not centralized registries of children with CP in the U.S., it is difficult to determine the extent to which the sample of children who participated in this project represents the

larger population of children with CP. It is possible that our sample reflects some type of self-selection bias. However, if the sample of children in the present study is representative of the larger population, our results suggest that the prevalence of speech and/or language impairment in children with CP is higher than previously estimated (76% in the present study vs. 60% reported by Bax et. al, 2006). To validate and extend the findings of the present study so that the full classification system described in this paper is examined, it is important that this work be replicated and scaled-up to include a larger number of children. Inclusion of children from countries where population characteristics are known would add international validity to the classification system and would enable determination of enrollment rates and the representativeness of the sample.

The proposed speech and language classification system is primarily oriented toward the identification and characterization of the constellation of speech and language *impairments* observed in children with CP. Accordingly, the measures we employed to differentiate among groups primarily reflected impairments. Consequently, the proposed classification system primarily targets the Body Structures and Functions domain of the World Health Organization's International Classification of Functioning, Disability, and Health (ICF). However, the argument could be made that gross judgments of *functional* speech abilities ("Activity" level of the ICF model) are also inherent in the proposed classification system (e.g. ability to produce speech) and measures of speech intelligibility and speech rate, used to differentiate among groups, are both indices of function ("Activity" level of the ICF model) (Yorkston et al., 1999a). Nonetheless, clearly lacking from this classification system is any indicator of participation, or of multimodal communication effectiveness. Additional tools designed to classify functional communication ability in individuals with CP are necessary and would complement the information provided by our speech and language classification system in important ways.

#### Acknowledgments

The authors thank the children with CP and their families who participated in this research, and the graduate and undergraduate students at the University of Wisconsin – Madison who assisted with data collection and data reduction. We also thank Gary Weismer and Ray Kent for helpful comments on earlier iterations of this manuscript. This study was funded by grants K23DC007114 and R01DC009411 from the National Institute on Deafness and Other Communication Disorders, National Institutes of Health (United States Department of Health and Human Services), and by a grant from the Cerebral Palsy International Research Foundation.

# References

- Achilles RF. Communicative anomalies of individuals with cerebral palsy. Cerebral Palsy Review 1955;16(5):15–24.
- Ansel BM, Kent RD. Acoustic-phonetic contrasts and intelligibility in the dysarthria associated with mixed cerebral palsy. Journal of Speech and Hearing Research 1992;35:296–308. [PubMed: 1573870]
- Bax M, Goldstein M, Rosenbaum P, Levinton A. Proposed definition and classification of cerebral palsy. Developmental Medicine & Child Neurology 2005;47(8):571–576. [PubMed: 16108461]
- Bax M, Tydeman C, Flodmark O. Clinical and MRI correlates of cerebral palsy: The European cerebral palsy study. Journal of American Medical Association 2006;296(13):1602–1608.
- Byrne M. Speech and language development of athetoid and spastic children. Journal of Speech and Hearing Research 1959;24(23):231–240.
- Carrow-Woolfolk, E. Test for Auditory Comprehension of Language. 3. Austin, TX: Pro-Ed; 1999.
- Cocozzelli C. Understanding canonical discriminant function analysis: Testing typological hypotheses. Journal of Social Service Research 1988;11:93–117.
- Cole KN, Mills PE. Agreement of language intervention triage profiles. Topics in Early Childhood Special Education 1997;17:119–130.

- Cole KN, Mills PE, Kelley D. Agreement of assessment profiles used in cognitive referencing. Language Speech and Hearing Services in the Schools 1994;25:25–31.
- Coplan J, Gleason JR. Unclear speech: Recognition and significance of unintelligible speech in preschool children. Pediatrics 1988;82:447–452. [PubMed: 3405680]
- Darley F, Aronson A, Brown J. Clusters of deviant speech dimensions in the dysarthrias. Journal of Speech and Hearing Research 1969;12:462–496. [PubMed: 5811846]
- Dickinson H, Parkinson K, Ravens-Sieberer G, Thyen U, Arnaud C, Beckung E, et al. Self-reported quality of life of 8–12 year old children with cerebral palsy: A cross-sectional European study. The Lancet 2007;369:2171–2178.
- Eliasson AC, Krumlinde-Sundholm L, Rosblad B, Beckung E, Arner M, Ohrvall AM, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. Developmental Medicine & Child Neurology 2006;48:549–554. [PubMed: 16780622]
- Fedrizzi E, Pagliano E, Andreucci E, Oleari G. Hand function in children with hemiplegic cerebral palsy: Prospective follow-up and functional outcome in adolescence. Developmental Medicine & Child Neurology 2003;45:85–91. [PubMed: 12578233]
- Fenson, L.; Resznick, S.; Thal, D.; Bates, E.; Hartung, J.; Reilly, J. The MacArthur Communicative Development Inventories. San Antonio, TX: Psychological Corporation; 1993.
- Gangil A, Patwari A, Aneja S, Ahuja B, Anand V. Feeding problems in children with cerebral palsy. Indian Pediatrics 2001;38:839–846. [PubMed: 11520994]
- Gorter JW, Rosenbaum PL, Hanna SE, Palisano RJ, Bartlett D, Russell D, et al. Limb distribution, motor impairment, and functional classification of cerebral palsy. Developmental Medicine & Child Neurology 2004;46:461–467. [PubMed: 15230459]
- Hanna SE, Law MC, Rosenbaum PL, King GA, Walter S, Pollock N, et al. Development of hand function among children with cerebral palsy: Growth curve analysis for ages 16 to 70 months. Developmental Medicine & Child Neurology 2003;45:448–455. [PubMed: 12828398]
- Hardy JC. Lung function of athetoid and spastic quadriplegic children. Developmental Medicine & Child Neurology 1964;6:378–388. [PubMed: 14210660]
- Higgins CM, Hodge MM. Vowel area and intelligibility in children with and without dysarthria. Journal of Medical Speech-Language Pathology 2002;10:271–277.
- Hodge, M.; Daniels, J. TOCS+ Intelligibility Measures. Edmonton, AB: University of Alberta; 2007.
- Hustad KC. Contribution of two sources of listener knowledge to intelligibility of speakers with cerebral palsy. Journal of Speech, Language, and Hearing Research 2007;50:1228–1240.
- Hustad KC, Lee J. Spectral and temporal changes in speech production associated with alphabet supplementation. Journal of Speech and Hearing Research 2008;51:1438–1450.
- Hustad KC, Sassano K. Effects of rate reduction on severe spastic dysarthria in cerebral palsy. Journal of Medical Speech-Language Pathology 2002;10:287–292.
- Irwin OC. Phonetic equipment of spastic and athetoid children. Journal of Speech and Hearing Disorders 1955;20:54–57. [PubMed: 14368749]
- Irwin OC. Correct status of vowels and consonants in the speech of children with cerebral palsy as measured by an integrated test. The Cerebral Palsy Journal 1968 January-February;:9–12. [PubMed: 5636108]
- Johnson K, Flemming E, Wright R. Response to Whalen et al. Language 2004;80:646–648.
- Johnson S, Marlow N, Wolke D, Davidson L, Marston L, O'Hare A, et al. Validation of a parent report measure of cognitive development in very preterm infants. Developmental Medicine and Child Neurology 2004;46:389–397. [PubMed: 15174530]
- Kennes J, Rosenbaum P, Hanna SE, Walter S, Russell D, Raina P, et al. Health status of school aged children with cerebral palsy: Information from a population based sample. Developmental Medicine & Child Neurology 2002;44:240–247. [PubMed: 11995892]
- Kent R, Forner L. Speech segment duration in sentence recitations by children and adults. Journal of Phonetics 1980;8(2):157–168.
- Kent, R.; Read, C. Acoustic Analysis of Speech. 2. Albany, NY: Singular/Thomson Learning; 2001.

- Kent, RD. Models of speech motor control: Implications from recent developments in neurophysiological and neurobehavioral science. In: Maassen, B.; Kent, RD.; Peters, HFM.; van Lieshout, PHHM.; Hulstijn, editors. Speech Motor Control in Normal and Disordered Speech. Oxford: Oxford University Press; 2004. p. 3-28.
- Kent RD, Weismer G, Kent JF, Vorperian HK, Duffy JR. Acoustic studies of dysarthric speech: Methods, progress, and potential. Journal of Communication Disorders 1999;32:141–186. [PubMed: 10382143]
- Kilbride HW, Thorstad K, Daily D. Preschool outcome of less than 801-gram preterm infants compared with full term siblings. Pediatrics 2004;113:742–747. [PubMed: 15060222]
- Kilincaslan A, Mukaddes N. Pervasive developmental disorders in individuals with cerebral palsy. Developmental Medicine and Child Neurology 2008;51:289–294. [PubMed: 19335564]
- Klatt D. Linguistic uses of segmental duration in English: Acoustic and perceptual evidence. Journal of Acoustical Society of America 1976;59:1208–1221.
- LeDorze G, Ouellet L, Ryalls J. Intonation and speech rate in dysarthric speech. Journal of Communication Disorders 1994;27:1–18. [PubMed: 8006203]
- Lee S, Potamianos A, Narayanan S. Acoustics of children's speech: Developmental changes of temporal and spectral parameters. Journal of Acoustical Society of America 1999;105:1455–1468.
- Leith WR, Steer MD. Comparison of Judged Speech Characteristics Of Athetoids and Spastics. Cerebral Palsy Review 1958:15–19.
- Lepage C, Noreau L, Bernard P, Fougeyrollas P. Profile of handicap situations in children with cerebral palsy. Scandinavian Journal of Rehabilitation Medicine 1998;30:263–272. [PubMed: 9825391]
- Light J, Collier B, Parnes P. Communicative interaction between young nonspeaking physically disabled children and their primary caregivers: Part I Discourse patterns. Augmentative and Alternative Communication 1985a;1:125–133.
- Light J, Collier B, Parnes P. Communicative interaction between young nonspeaking physically disabled children and their primary caregivers: Part II Modes of communication. Augmentative and Alternative Communication 1985b;1:125–133.
- Light J, Collier B, Parnes P. Communicative interaction between young nonspeaking physically disabled children and their primary caregivers: Part III -Communicative Function. Augmentative and Alternative Communication 1985c;1:98–107.
- Liptak GS, O'Donell M, Conaway M, Chumlea WC, Worley G, Henderson RC, et al. Health status of children with moderate to severe cerebral palsy. Developmental Medicine & Child Neurology 2001;43:364–370. [PubMed: 11409824]
- Liu HM, Tsao FM, Kuhl PK. The effect of reduced vowel working space on speech intelligibility in Mandarin-speaking young adults with cerebral palsy. Journal of Acoustical Society of America 2005;117(6):3879–3889.
- Milenkovic, P. TF32. Madison, WI: University of Wisconsin Madison; 2002.
- Morris C, Galuppi BE, Rosenbaum PL. Reliability of family report for the Gross Motor Function Classification System. Developmental Medicine & Child Neurology 2004;46:455–460. [PubMed: 15230458]
- Morris C, Kurinczuk JJ, Fitzpatrick R, Rosenbaum PL. Reliability of the Manual Ability Classification System for children with cerebral palsy. Developmental Medicine & Child Neurology 2006a; 48:950–953. [PubMed: 17109781]
- Morris C, Kurinczuk JJ, Fitzpatrick R, Rosenbaum PL. Who best to make the assessment? Professionals' and families' classifications of gross motor function in cerebral palsy are highly consistent. Archives of Disease in Childhood 2006b;91:675–679. [PubMed: 16638783]
- Odding E, Roebroeck ME, Stam HJ. The epidemiology of cerebral palsy: Incidence, impairments and risk factors. Disability and Rehabilitation 2006;28(4):183–191. [PubMed: 16467053]
- Palisano R, Hanna SE, Rosenbaum P, Russell D, Walter S, Wood E, et al. Validation of a model of gross motor function for children with cerebral palsy. Physical Therapy 2000;80(10):974–985. [PubMed: 11002433]

- 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. Developmental Medicine and Child Neurology 1997a;39:214–223. [PubMed: 9183258]
- 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. Developmental Medicine & Child Neurology 1997b;39:214–223. [PubMed: 9183258]
- Pan BA, Rowe MA, Spier E, Tamis-LeMonda C. Measuring productive vocabulary of toddlers in lowincome families: Concurrent and predictive validity of three sources of data. Journal of Child Language 2004;3:587–608. [PubMed: 15612391]
- Paneth N, Hong T, Korzeniewski S. The Descriptive Epidemiology of Cerebral Palsy. Clinics in Perinatology 2006;33:251–267. [PubMed: 16765723]
- Pennington L, McConachie H. Interaction between children with cerebral palsy and their mothers: The effects of speech intelligibility. International Journal of Language and Communication Disorders 2001a;36:371–393. [PubMed: 11491485]
- Pennington L, McConachie H. Predicting patterns of interaction between children with cerebral palsy and their mothers. Developmental Medicine & Child Neurology 2001b;43:83–90. [PubMed: 11221909]
- Platt LJ, Andrews G, Young M, Quinn PT. Dysarthria of adult cerebral palsy: I. Intelligibility and articulatory impairment. Journal of Speech and Hearing Research 1980;22:28–40. [PubMed: 7442182]
- Reilly S, Skuse D, Poblete X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: A community survey. The Journal of Pediatrics 1996;129(6):877–882. [PubMed: 8969730]
- Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M. A report: The definition and classification of cerebral palsy. Developmental Medicine & Child Neurology 2007;49(s108):8–14.
- Rosenbaum PL, Walter SD, Hanna SE, Palisano RJ, Russell DJ, Raina P, et al. Prognosis for gross motor function in cerebral palsy: Creation of motor development curves. Journal of the American Medical Association 2002;288:1357–1363. [PubMed: 12234229]
- Rutherford BR. Frequency of articulation substitutions in children handicapped by cerebral palsy. Journal of Speech and Hearing Disorders 1939;4:285–287.
- Rutherford BR. A comparative study of loudness, pitch, rate, rhythm and quality of the speech of children handicapped by cerebral palsy. Journal of Speech Disorders 1944;9:263–271.
- Sigurdardottir S, Eriksdottir A, Gunnarsdottir E, Meintema M, Arnadottir U, Vik T. Cognitive profile in young Icelandic children with cerebral palsy. Developmental Medicine and Child Neurology 2008:50.
- Smith, A.; Goffman, L. Interaction of motor and language factors in the development of speech production. In: Maassen, B.; Kent, RD.; Peters, HFM.; van Lieshout, PHHM.; Hulstijn, W., editors. Speech Motor Control in Normal and Disordered Speech. Oxford: Oxford University Press; 2004. p. 225-252.
- Smith A, Goffman L, Stark RE. Speech motor development. Seminars in Speech and Language 1995;16(2):87–99. [PubMed: 7621336]
- Smith A, Zelaznik HN. Development of functional synergies for speech motor coordination in childhood and adolescence. Developmental Psychobiology 2004;45:22–33. [PubMed: 15229873]
- Smith B. Temporal aspects of English speech production: A developmental perspective. Journal of Phonetics 1978;6:37–68.
- Smith B, Kenney M. An assessment of several acoustic parameters in children's speech production development: longitudinal data. Journal of Phonetics 1998;26:95–108.
- Strand, EA. The integration of speech motor control and language formulation in process models of acquisition. In: Chapman, RS., editor. Processes in Language Acquisition and Disorders. St. Louis, Baltimore, Boston, Chicago, London, Philadelphia, Sydney, Toronto: Mosby; 1992. p. 86-107.
- Turner G, Tjaden K, Weismer G. The influence of speaking rate on vowel space and speech intelligibility for individuals with amyotrophic lateral sclerosis. Journal of Speech and Hearing Research 1995;38:1001–1013. [PubMed: 8558870]

- Vorperian H, Kent R. Vowel acoustic space development in children: A synthesis of acoustic and anatomical data. Journal of Speech, Language, and Hearing Research 2007;50:1510–1545.
- Walker JF, Archibald L, Cherniak S, Fish V. Articulation rate in 3- and 5- year old children. Journal of Speech and Hearing Research 1992;35:4–13. [PubMed: 1735975]
- Weismer G, Jeng J, Laures JS, Kent R. Acoustic and intelligibility characteristics of sentence production in neurogenic speech disorders. Folia Phoniatrica 2001;53:1–18.
- Weismer G, Laures JS, Jeng J, Kent RD. Effect of speaking rate manipulations on acoustic and perceptual aspects of the dysarthria in amyotrophic lateral sclerosis. Folia Phoniatrica et Logopaedica 2000;52:201–219. [PubMed: 10965174]
- Weismer, G.; Martin, R. Acoustic and perceptual approaches to the study of intelligibility. In: Kent, R., editor. Intelligibility in Speech Disorders. Philadelphia: John Benjamins Publishing Co; 1992. p. 67-118.
- Wood E, Rosenbaum P. The gross motor classification system for cerebral palsy A study of reliability and stability over time. Developmental Medicine & Child Neurology 2000;42:292–296. [PubMed: 10855648]
- Workinger, M.; Kent, R. Perceptual analysis of the dysarthria in children with athetoid and spastic cerebral palsy. In: Moore, C.; Yorkston, K.; Beukelman, D., editors. Dysarthria and apraxia of speech: Perspectives on management. Baltimore: Paul H. Brookes; 1991. p. 109-126.
- Yeargin-Allsopp M, Braun K, Doernbery N, Benedict R, Kirby R, Durkin M. Prevalence of cerebral palsy in 8-year-old children in three areas of the United States in 2002: A multisite collaboration. Pediatrics 2008;121:547–554. [PubMed: 18310204]
- Yorkston, KM.; Beukelman, DR.; Strand, E.; Bell, K. Management of Motor Speech Disorders in Children and Adults. 2. Austin, TX: Pro-Ed; 1999a.
- Yorkston, KM.; Beukelman, DR.; Strand, EA.; Bell, KR. Management of motor speech disorders in children and adults. 2. Austin, TX: Pro Ed; 1999b.
- Zimmerman, I.; Steiner, V.; Pond, R. Preschool Language Scale-4. 4. San Antonio, TX: Psychological Corporation; 2002.

# Appendix A

Decision making rubric for assigning children into a priori communication profile groups based on clinical impressions from the data collection session.







Schematic model of hypothesized communication profile groups.







Schematic model of communication profile groups examined in this study.





#### Figure 3.

Vowel space area by Profile Group. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Note that children in Profile Group ANAR (all of whom were unable to speak) did not contribute vowel space data. Vowel area is measured in Hz<sup>2</sup>



#### Figure 4.

Speech rate by profile group. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Note that children in Profile Group ANAR (all of whom were unable to speak) did not contribute speech rate data. Rate data are measured in syllables per minute and are based on the duration of recited utterances, including all pauses.



#### Figure 5.

Speech intelligibility ratings by profile group. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Profile Group ANAR refers to children who were anarthric. Intelligibility ratings are based on a 7-point likert scale where 1 = difficult or impossible to understand and 7 = very easy to understand.

Language Comprehension Score by Profile Group



#### Figure 6.

Language comprehension scores by profile group. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Profile Group ANAR refers to children who were anarthric. Standard language scores are based on a mean of 100 and a standard deviation of 15.





Description of hypothesized communication profile groups examined in this study.

| Group   | Description   |
|---------|---|
| NSMI    | No evidence of speech motor impairment based on clinical assessment; language abilities age appropriate (typically developing) or impaired based on clinical assessment |
| SMI-LCT | Evidence of speech motor impairment; language abilities age appropriate (typically developing) based on clinical assessment   |
| SMI-LCI | Evidence of speech motor impairment; language abilities impaired based on clinical assessment   |
| ANAR    | Unable to produce functional speech (anarthria); language abilities age appropriate (typically developing), impaired, or unknown based on clinical assessment.          |

Demographics of children with CP including chronological age (CA), sex ratio, type of CP, and Gross Motor Function Classification System level (GMFCS) (Palisano et al., 1997a) by hypothesized communication profile group.

|                            | Group NSMI (n = 8) | Group SMI-LCT (n = 9) | Group SMI-LCI (n = 6) | Group ANAR (n = 11) |
|----------------------------|--------------------|-----------------------|-----------------------|---------------------|
| Mean Age (SD)              | 54.7 (1.1)         | 53.5 (1.7)            | 54.9 (1.8)            | 54.7 (2.1)          |
| Male: female ratio         | 5:3                | 2:7                   | 3:3                   | 8:3                 |
| Type of CP                 |                    |                       |                       |                     |
| Diplegia                   | 4                  | 2                     | 1                     | 0                   |
| Hemiplegia (left)          | 3                  | 1                     | 0                     | 0                   |
| Hemiplegia (right)         | 0                  | 1                     | 1                     | 0                   |
| Quadriplegia               | 0                  | 2                     | 2                     | 8                   |
| Dyskinesia                 | 0                  | 2                     | 0                     | 0                   |
| Ataxia                     | 0                  | 0                     | 1                     | 0                   |
| Mixed                      | 0                  | 0                     | 1                     | 2                   |
| Unknown                    | 1                  | 1                     | 0                     | 1                   |
| GMFCS level                |                    |                       |                       |                     |
| Level I                    | 2                  | 1                     | 0                     | 0                   |
| Level II                   | 6                  | 4                     | 2                     | 1                   |
| Level III                  | 0                  | 2                     | 2                     | 0                   |
| Level IV                   | 0                  | 2                     | 0                     | 4                   |
| Level V                    | 0                  | 0                     | 2                     | 6                   |
| Cortical visual impairment | 0                  | 0                     | 0                     | 3                   |
| Corrected vision           | 0                  | 2                     | 4                     | 5                   |

Follow-up tests examining comparisons between profile groups on each of the dependent measures. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Profile Group ANAR refers to children who are anarthric.

| Contrast               | Mean difference | Z     | р     |
|------------------------|-----------------|-------|-------|
| Vowel area             |                 |       |       |
| NSMI vs. SMI-LCT       | 36164.10        | -2.12 | .036* |
| NSMI vs. SMI-LCI       | 61323.80        | -2.97 | .001* |
| SMI-LCT vs. SMI-LCI    | 25159.70        | -1.77 | .080  |
| Speech rate            |                 |       |       |
| NSMI vs. SMI-LCT       | 66.89           | -2.89 | .002* |
| NSMI vs. SMI-LCI       | 65.03           | -2.71 | .005* |
| SMI-LCT vs. SMI-LCI    | -1.87           | -0.47 | .689  |
| Intelligibility        |                 |       |       |
| NSMI vs. SMI-LCT       | 2.67            | -2.71 | .006* |
| NSMI vs. SMI-LCI       | 2.58            | -2.93 | .003* |
| NSMI vs. ANAR          | 4.90            | -3.97 | .001* |
| SMI-LCT vs. SMI-LCI    | 08              | -0.24 | .864  |
| SMI-LCT vs. ANAR       | 2.24            | -3.23 | .003* |
| SMI-LCI vs. ANAR       | 2.33            | -3.62 | .001* |
| Language comprehension |                 |       |       |
| NSMI vs. SMI-LCT       | 2.97            | -0.83 | .423  |
| NSMI vs. SMI-LCI       | 38.08           | -3.12 | .001* |
| NSMI vs. ANAR          | 54.57           | -3.73 | .001* |
| SMI-LCT vs. SMI-LCI    | 35.11           | -3.19 | .001* |
| SMI-LCT vs. ANAR       | 51.59           | -3.85 | .001* |
| SMI-LCI vs. ANAR       | 16.48           | -2.71 | .007* |

statistical significance at p<.05

children with speech motor involvement and age appropriate (typically developing) language skills; Profile Group SMI-LCI refers to children with speech motor involvement and language impairment. Profile Group ANAR refers to children who were anarthric. CDFA refers to canonical discriminant Comparisons among classification methods. Profile Group NSMI refers to children with no speech motor involvement; Profile Group SMI-LCT refers to function analysis

| Classification method comparison                                    | Percent agreement | Number of disagreements | Nature of disagreement                    |
|---|-------------------|-------------------------|---|
| A priori assignment & CDFA  | 97%               | 1                       | NSMI vs. SMI-LCT (child 3)                |
| A priori assignment & CDFA cross-validated                          | 91%               | 3                       | NSMI vs. SMI-LCT (child 3, 20)            |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 18)            |
| A priori assignment & Cluster analysis                              | 79%               | 7                       | NSMI vs. SMI-LCT (child 4, 16, 30)        |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 9, 23, 25)     |
|   |                   |                         | SMI-LCT vs. ANAR (child 18)               |
| CDFA & CDFA cross-validated   | 94%               | 2                       | NSMI vs. SMI-LCT (child 20)               |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 18)            |
| CDFA & Cluster analysis   | 76%               | 8                       | NSMI vs. SMI-LCT (child 3, 4, 16, 30)     |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 9, 23, 25)     |
|   |                   |                         | SMI-LCT vs. ANAR (child 18)               |
| CDFA-cross validated & Cluster analysis                             | 74%               | 6                       | NSMI vs. SMI-LCT (child 3, 4, 16, 20, 30) |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 9, 23, 25)     |
|   |                   |                         | SMI-LCI vs. ANAR (child 18)               |
| A priori assignment, CDFA, CDFA cross-validated, & Cluster analysis | 74%               | 6                       | NSMI vs. SMI-LCT (child 3, 4, 16, 20, 30) |
|   |                   |                         | SMI-LCT vs. SMI-LCI (child 9, 23, 25)     |
|   |                   |                         | SMI-LCT vs. SMI-LCI vs. ANAR (child 18)   |