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Vocal characteristics of children with cerebral palsy and anarthria

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Abstract

Purpose: This study aimed to investigate the vocal characteristics of children with CP and anarthria using the stage model of vocal development.

Method: Vocal characteristics of 39 children with CP and anarthria around four years of age were analyzed from laboratory-based caregiver-child interactions. Perceptual coding analysis was conducted using the *Stark Assessment of Early Vocal Development-Revised* to examine vocal complexity, volubility, and consonant diversity.

Results: Children predominately produced vocalizations corresponding to the two earliest stages of vocal development characterized by vowel-like utterances. They showed a limited attainment of consonantal features with low consonant diversity and variably low vocal rates.

Conclusion: Our results demonstrate that underlying neurological impairments resulting in an anarthric status in children with CP affect the progression of speech motor development and their ability to advance beyond early vocal stages. These findings highlight the importance of considering alternative communication modalities for children demonstrating similar vocal characteristics beyond expected periods of development.

Keywords: speech; nonverbal; vocalization

Vocal characteristics of children with cerebral palsy and anarthria

Over 80% of children with cerebral palsy (CP) also have speech motor involvement, including approximately 25% with *anarthria*, defined as an absence of speech due to substantially impaired neurological functioning of the speech motor system (Mei et al., 2020; Nordberg et al., 2013). Anarthria in children with CP can result in social isolation, loneliness, and reduced quality of life which highlights the critical need to enhance alternative communication modalities (Currie & Szabo, 2020; Kwan et al., 2020; Lindsay & McPherson, 2012; Mei et al., 2014; Smith & Hustad, 2015). The ability to predict which children with CP will ultimately have anarthria is currently not possible, and a "wait and see" approach prevails, which may delay delivery of the best speech and language treatments (i.e. comprehension voice output augmentative and alternative communication (AAC) interventions) for these children.

There is a growing body of research aiming to improve the early prediction of speech motor involvement in children with CP. Studies suggest that the age at which children are able to produce elicited single word approximations has a significant impact on the rate of intelligibility development and on utterance length outcomes at four years of age (Hustad et al., 2019, 2020). Specifically, children who produced words at earlier ages had better outcomes than those who produced words later in development. Moreover, approximately 73% of children who were unable to speak at two years were later classified as anarthric at four years (Hustad et al., 2017). Studies also indicate that early speech intelligibility is highly predictive of later intelligibility at older ages (Hustad et al., 2023; Mahr et al., 2020). Children with CP and no functional speech by four years of age tend to remain severely impaired or anarthric, while those with some functional speech at four years exhibit continued speech development through 10 years of age (Long, Mahr, et al., 2022). Despite these findings, predicting speech motor involvement including anarthria remains challenging, particularly at pre-linguistic or early ages.

CP is unique from many other neurodevelopmental conditions (e.g., autism, Down syndrome) due to its etiology, which arises from nonprogressive disturbances that affect neuromotor development in the fetal or infant brain (Rosenbaum et al., 2007). Although the underlying neuropathology is generally stable for CP, children nonetheless are in a constant state of change owing to the impacts of development which can have complex interactions with neuropathology. Unlike gross and fine motor limitations which can be observed in infancy and often trigger a diagnostic assessment for CP (Novak et al., 2017), speech motor impairments like anarthria cannot be diagnosed until speaking ages in early childhood (i.e., after the age of two years). Although gross motor and speech motor functioning are highly correlated (Choi et al., 2018; Monbaliu et al., 2017), predicting any level of speech motor involvement in infancy is still not possible.

The infraphonological stage model of infant vocal development (Oller, 2000) has potential to offer insights into the identification of early markers of speech motor involvement in

children with CP. This model is based in the longstanding assumption that the emergence of five robust stages of infant vocal development reflects a canalized pattern of phonatory and articulatory parameters leading to the production of first words, wherein each stage is considered foundational to the next (Koopmans-van Beinum & van der Stelt, 1986; Nathani et al., 2006; Oller, 1978; Stark, 1980). Specifically, infants produce reflexive vocalizations (e.g., grunts, cries) shortly after birth, followed by the production of "gurgly" cooing vocalizations, then vocalizations with imprecise (i.e., marginal) consonant-vowel (CV) syllables and vocal play with pitch and amplitude, and eventually the production of syllables with adult-like (i.e., canonical) CV syllables by the second half year of life. The successful emergence of these stages is considered a manifestation of the achievement of all necessary parameters (phonation, vocal posturing, full nuclei resonance, and rapid CV formant transitions) to produce syllables for speech by the end of the first year.

Previous research has shown that infants later diagnosed with communication impairments often exhibit delays in the emergence of vocal stages, particularly canonical babbling (Lang et al., 2019; Lohmander et al., 2017; Lynch et al., 1995; Masataka, 2001; Oller et al., 1999; Overby et al., 2020; Yankowitz et al., 2022). However, few studies have explicitly examined earlier vocal stages in the context of speech motor disorders (Long, Christensen, et al., 2023).

Given the etiology of CP, we expect that specific markers of speech motor involvement would be present from birth—regardless of the age of formal CP medical diagnosis. We hypothesize that one form an early marker of speech motor involvement may take is disruption in the canalization process of vocal forms as described in the stage model of vocal development. Several studies with small samples have shown evidence of delays in canonical babbling and higher rates of marginal syllables at and beyond 12 months of age (Levin, 1999; Long, Eichorn, et al., 2023; Long & Hustad, 2023; Nyman & Lohmander, 2018; Ward et al., 2022, 2023). However, the scant body of evidence limits our current capacity to prospectively predict differential levels of speech motor involvement, including anarthria.

A possible avenue to address this gap is to retrospectively examine the vocal characteristics of young children with CP and anarthria with similar vocal analysis methods used for prelinguistic infants. This approach would allow us to establish the range and potential limits of the vocal development of children known to have anarthria. Generally, studies suggest that children with CP who are able to produce speech show delayed speech development time frames in terms of the age at which steepest intelligibility growth occurs, and protracted time frames of growth (Hustad et al., 2020; Long, Mahr, et al., 2022; Mahr et al., 2020). Similar patterns may exist in the vocal development time frames of children with anarthria; further, studies have not examined the limits of vocal development in these children. Such information could provide an important starting point for identifying vocal features of children who will not develop functional speech, which may inform early identification of anarthria in future studies.

The research objective of the present study was to characterize vocal complexity, volubility, and consonant diversity of children with anarthria and CP at speaking ages. Specifically, we sought to examine the limits of vocal development at 4 years of age in children who are unable to produce functional speech due to severe speech motor impairment (anarthria). We hypothesized that children with CP and anarthria would exhibit: 1) high rates of vocalizations corresponding to earlier stages of infant vocal development, 2) low vocal rates, and 3) small consonant inventories.

Method

This study was approved by the institutional review board for social and behavioral sciences at the University of Wisconsin-Madison (IRB #2018-0580). Written consent was obtained from caregivers prior to participation.

Participants

Thirty-nine children with anarthria and CP around four years of age (mean = 50.8 months, SD = 1.7) were included in the present study. Participating children were selected from a larger longitudinal cohort (n = 139) examining speech and communication development in children with CP. All children in the longitudinal project had a medical diagnosis of CP.

The inclusion criteria for the present study were 1) a classification of anarthria (further elaborated below), and 2) a laboratory visit with a caregiver-child interaction between 48-54 months of age. The longitudinal database included forty-nine children previously classified as anarthric. Four were excluded because they did not have a laboratory visit within the selected age band. Two were excluded because the caregiver-child interaction session was too short (<5 minutes). One was excluded because they did not have a diagnosis of CP. Three were excluded because they produced <5 vocalizations in the recording for analysis. Thirty-nine children therefore qualified for the present study. No child was excluded on the basis of co-occurring conditions. The four-year age band was selected as an approximate age at which speech impairments are commonly diagnosed in children with CP and no longer constitute as a delay.

Table 1 presents the participant demographics. Racial demographics are representative of the recruitment region of Madison, Wisconsin. Gross motor and fine motor functioning was reported by caregivers using the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997) and the Manual Abilities Classification System (MACS; Eliasson et al., 2006), respectively. Each system describes gross motor or fine motor functioning levels, respectively, using an ordinal rating scale from no functional impairment (I) to substantially reduced functioning (V).

Table 1. Participant demographics

Variable	N = 39
Average age (SD)	50.8 months (1.7)
Sex	
Male	24
Female	15
Race/Ethnicity	
White or Caucasian	35
Black or African American	2
Hispanic or Latine	1
White and Asian	1
CP Type	
Spastic	23
Mixed	5
Hypotonic	2
Not reported	9
GMFCS Level	
I	0
II	6
III	1
IV	6
V	18
Not reported	8
MACS Level	
I	1
II	5
III	7
IV	18
V	8
Co-occurring medical conditions	
Seizure history	29
Cortical visual impairment (CVI)	16
Other non-CVI visual impairment	14
Genetic syndrome	7
Brain malformation	3
Cardiovascular disorder	2
Autism	2
Hearing impairment	2
Respiratory disorder	2
None reported	3

Anarthria Classification. All children participating in the larger longitudinal project were previously classified into one of four Speech-Language Profile Groups (SLPG, Hustad et al., 2010) to describe their speech and language performance using speech and language assessment tasks (Hustad et al., 2017, 2018; McFadd & Hustad, 2013). Children received a designation of anarthria if they produced fewer than five words or word approximations based on caregiver-report and clinical observation during laboratory sessions.

Table 2 describes group-level speech and language characteristics of the 39 children. Speech therapy enrollment and augmentative and alternative communication (AAC)-targeted intervention information were caregiver-reported at the time of the laboratory visit.

Table 2. Speech therapy and language comprehension

Variable	N = 39
Enrolled in speech therapy	
Yes	35
No	4
Speech therapy targeting AAC	
Yes	14
No	25
Language Comprehension	
Median age equivalency (range)	7 mo (3-51 mo)

Recording Material

Child vocal characteristics were examined during laboratory-based, caregiver-child interaction sessions, with a mean duration of 13.7 minutes (SD = 3.9). During these sessions, caregivers were asked to interact and speak with their child naturally using toys or engaging with interactive books. Sessions were extracted from full laboratory visits lasting 1- to 1.5 hours in length that included other tasks such as caregiver interviews and standardized and informal speech and language assessments.

Coding Procedure

All coding was conducted using the *Action Analysis Coding Training* behavioral coding software (AACT; Delgado & Oller, 1999). Using this program, two undergraduate research assistants classified child vocalizations according to the *Stark Assessment of Early Vocal Development-Revised* (SAEVD-R, Nathani et al., 2006). One primary coder conducted all coding for all children; the secondary coder served as the reliability coder. Both coders received

extensive vocal coding training from the first author using training video modules and regular feedback sessions on practice coding material. Formal vocal coding by students began only after their training reliability with the instructor (first author) exceeded 85% during training. Both coders were blind to the age of children and the hypotheses of this study; however, neither coder was blind to the anarthria classification of children. The SAEVD-R classifies child vocalizations across 23 categories within five hierarchical levels of developmental complexity according to the emergent trajectory of articulatory complexity for typically developing infants (Table 3).

Table 3. Stark Assessment of Early Vocal Development-Revised (Nathani et al. 2006)

Level	Expected onset age	Vocalization types
Level 1: Reflexive	0-2 mo	Vegetative noises (burp, cough, etc.), crying, fussing, short grunt-like vocalizations with muffled resonance
Level 2: Control of Phonation	1-4 mo	Vowel-like (i.e., quasivowel) vocalizations that are not fully resonant, closants, raspberries, trills, clicks, laughs
Level 3: Expansion	3-8 mo	Fully resonant vowels, glides, ingresses, squeals, marginal CV syllables with slow formant transitions and imprecise consonant-like features
Level 4: Canonical Syllables	5-10 mo	Canonical CV syllables with rapid formant transition and precise consonantal features in single, reduplicated, and variegated combinations; whispers, and CVC or CVCV syllable structures
Level 5: Advanced Forms	9-18 mo	Complex, multisyllabic strings (e.g., VC, CCV, VCVC), canonical utterances with varied intonation patterns (i.e., jargon), diphthongs with rapid vowel formant transitions

A single level was assigned to each utterance produced by children according to the highest level of developmental complexity observed across all syllables within the utterance. For example, an utterance containing a marginal and canonical syllable would be classified as Level 4 because it contained a canonical syllable, which is a higher level of complexity than a marginal syllable. *Utterances* were separated according to the "breath group criterion" defined as an audible ingress or pauses that could include ingressive breaths (Lynch et al., 1989; Nathani & Oller, 2001; Stark, 1980). Coders also marked syllable-level consonants for all utterances classified at Level 4 or Level 5 containing a mature canonical syllable. A total count of utterances classified within each level and the number of different consonants for each child was obtained to calculate vocal complexity, volubility, and consonant diversity measures described below.

Vocal Measures

Vocal Complexity: Level ratios for each of the five developmental levels of complexity were calculated as the number of utterances classified within each level divided by the total number of all utterances produced by each child. Following the SAEVD-R coding scheme, non-speech-like vocalizations (i.e., vegetative noises, cries, and laughs) were coded but excluded from analysis (Nathani et al., 2006). Five level ratios (Level 1, Level 2, Level 3, Level 4, Level 5) of speech-like vocalizations were thus calculated for each child to reflect their proportion of vocalizations produced within each vocal complexity level. The largest ratio across the five levels for each child was calculated as their *Highest Ratio,* a measure of each child's most frequent vocal type across the five levels. The highest level with a ratio $\geq$0.15 was calculated for each child as their *Established Level*. The original SAEVD-R paper reported that the Highest Ratio for typically developing children between 9-15 months was at Level 3, and Level 4 by 16-20 months. The Established Level ($\geq$ 0.15) for children between 9-15 months was Level 4, and between 16-20 months was Level 5 (Nathani et al., 2006). For reference, typically developing children above 30 months have demonstrated canonical proportions >0.40 (Cychosz et al., 2021; Hitczenko et al., 2023), corresponding to Levels 4 and 5 on the SAEVD-R.

Volubility: *Vocal Rate* was calculated as the number of utterances per minute. The total number of utterances was tabulated and divided by the duration in minutes of each child's interaction recording. Prior work has established in typical development an average vocal rate of 4-5 utterances per minute for infants between 0-12 months (Oller et al., 2019). Also for reference, typically developing 4-year-old children produce four to six word sentences with ~75% speech intelligibility at a rate of ~3 syllables per second (ASHA, 2023; CDC, 2023; Hustad et al., 2021; Mahr et al., 2021).

Consonant Diversity: A *Consonant Inventory* was calculated for each child as the total number of different true consonants produced at least once. Vihman et al. (1985) defines a true consonant as supraglottal articulatory constrictions, excluding glides and glottal stops, that is perceived as intentionally produced by the child. Prior work has established an inventory of 6-8 acquired consonants in typically developing children by the end of the first year (Morgan & Wren, 2018). We acknowledge the inherent challenge in conducting a phonetic analysis for children with anarthria (many of whom also had substantially reduced language and cognitive abilities); thus, the present paper centers on consonant diversity as a reflection of the range of articulatory posturing observed within our sample.

Reliability

We randomly selected 10 recordings each (24%) for inter-rater reliability analysis using the intraclass correlation coefficient (ICC, Shrout & Fleiss, 1979) with descriptive interpretations from Koo & Li (2016). We used a single score, absolute agreement, two-way random effects model and found good reliability between the two raters for level ratios, ICC (2, 1) = 0.83, 95%

CI [.71, .90], $p < .001$, excellent reliability for vocal rate, ICC (2, 1) = 0.97, 95% CI [.84, .99], $p < .001$, and moderate reliability for consonant inventories, ICC (2, 1) = 0.69, 95% CI [.29, .89], $p = .002$.

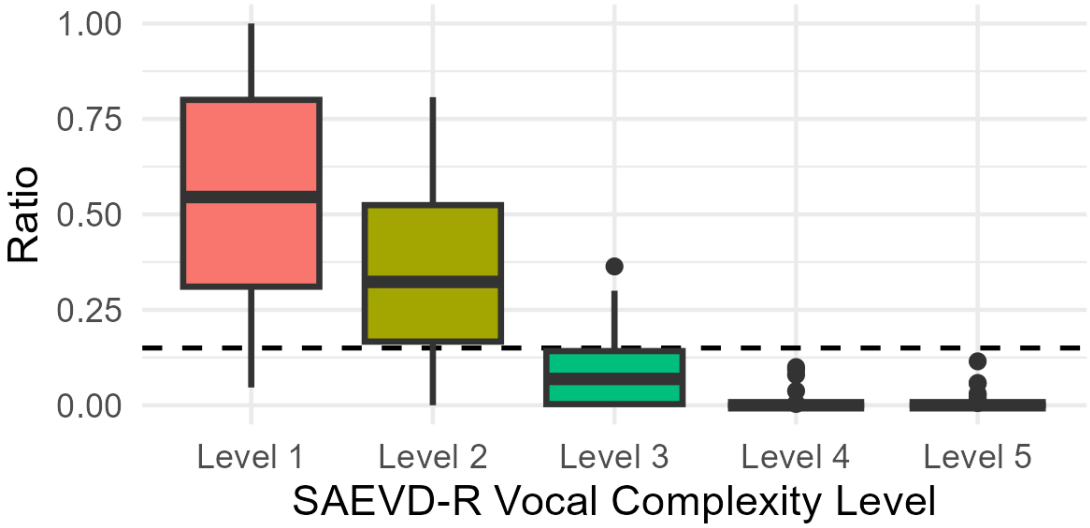
Results

Vocal complexity, volubility, and consonant diversity of the 39 children in our sample are described below. From these children, a total of 2,841 utterances were identified and classified using the SAEVD-R coding scheme. Following Ertmer et al. (2007), SAEVD-R level ratios were interpreted using the following criteria: "very high" ratios were ≥ 0.60, "high" ratios were between 0.40—0.59, "moderate" ratios were between 0.20—0.39, "low" ratios were between 0.10—0.19, and "very low" ratios were ≤ 0.09.

Vocal complexity

Figure 1 presents the distribution of vocal developmental complexity level ratios calculated across the five SAEVD-R levels. At the group level, our sample had a high median Level 1 ratio of 0.55 (range: 0.05-1.00) and a moderate median Level 2 ratio of 0.32 (range: 0.00-0.81). They had very low median Level 3 ratios: 0.07 (range: 0.00-0.36), Level 4 ratios: 0.00 (range: 0.00-0.10), and Level 5 ratios: 0.00 (range: 0.00-0.12).

Figure 1. Ratio distribution by vocal complexity level



Note. Ratio distributions across the five vocal complexity levels for 39 children with CP and anarthria. The dotted line indicates the 0.15 criterion representative of vocal stage attainment. SAEVD-R = Stark Assessment of Early Vocal Development-Revised.

Highest Ratio. Level 1 was the Highest Ratio level for 24 children (62%). Level 2 was the Highest Ratio level for 15 children (38%). No children (0%) had a Highest Ratio at Level 3, Level 4, or Level 5. These findings suggest a preponderance of vocalizations corresponding to the Reflexive (Level 1) and Control of Phonation (Level 2) stages of vocal development produced by children.

Established Level: For the 39 children who produced ≥ 5 vocalizations, Level 1 was the Established Level for eight children (21%). Level 2 was the Established Level for 22 children (56%). Level 3 was the Established Level for nine children (23%). No children (0%) had an Established Level at Level 4 or Level 5.

These findings indicate that most children in our sample had attained the Reflexive (Level 1) or Control of Phonation (Level 2) stages of vocal development. Several children had attained the Expansion (Level 3) stage by four years; however no child had a highest ratio level at Level 3 or higher.

Volubility

The mean vocal rate of children was 5.23 utterances per minute (SD = 4.40), with a range of 0.49 to 22.39 utterances per minute.

Consonant Diversity

Eleven (28%) children produced at least one true consonant. Within this subsample, one child (3%) produced three different true consonants, one child (3%) produced two different true consonants, and nine children (23%) produced one true consonant. Twenty-eight children (72%) did not produce any true consonants. The overall inventory of true consonants produced across the 11 children was [b], [d], [t], [g], [n]. Of these, [b] was the most common consonant, represented in the consonant inventory of six out of the 11 children (55%); [n] was represented in the consonant inventory of three children (27%). Finally, [d] and [g] were represented in the consonant inventory of two children each (18%), and [t] was represented in the consonant inventory of one child (9%).

Discussion

The present study aimed to characterize vocal complexity, volubility, and consonant diversity of children with anarthria and CP at approximately four years of age. Following the stage model of vocal development (Oller, 2000), we found that these children predominately produced vowel-like vocalizations corresponding to the two earliest stages of vocal development, with very limited ability to produce consonant-like constrictions in the vocal tract.

We selected vocal measures based on a scoping review of work examining vocal characteristics of infants at risk for speech motor involvement (Long, Christensen, et al., 2023) wherein vocal ratios, vocal rates, and consonant inventories were the most common measures reported. The SAEVD-R was used as a detailed coding scheme which provided a normative framework for typical vocal stage emergence (Nathani et al., 2006; Stark, 1980, 1981). This measure was selected over the commonly used binary coding classification of canonical vs non-canonical syllables in utterances (Long, Eichorn, et al., 2023; Long, Ramsay, et al., 2022; Nyman et al., 2021; Oller et al., 2019) to support a more comprehensive analysis of articulatory development in children with CP and anarthria. This is notable because descriptive classifications of anarthria are based on the absence of speech at speaking ages (Hustad et al., 2010; Mei et al., 2020); thus, this study presents a novel approach to quantifying vocal characteristics of anarthria for the purposes of informing its prediction.

The 0.15 criterion was used to indicate vocal stage attainment of children's Established Level; however, we note that the developers of the SAEVD-R utilized a 0.10 criterion because "it was expected that trained listeners would identify the emergence of new vocal types earlier than parents" (Nathani et al., 2006, p. 358). Recent work using the SAEVD-R has used the 0.10 criterion given this suggestion from the developers (Ward et al., 2023). However, this lower criterion has yet to be validated to the same extent as the 0.15 criterion (Nyman et al., 2021; Nyman & Lohmander, 2018; Oller et al., 1998). Thus, the present study retained the more rigorous 0.15 criterion as the standard cut-off.

The majority of children had a Highest Ratio at Level 1 (62%), corresponding to the Reflexive stage of vocal development, but had an Established Level at Level 2 (56%), corresponding to the Control of Phonation stage of vocal development. A preponderance of utterances at these two earliest stages of developmental complexity highlights the substantially reduced speech motor control affecting the ability to execute rapid movements and articulatory posturing for consonants. Despite this, nine children in our sample (23%) had an Established Level at Level 3 corresponding to the Expansion Stage, reflecting some mastery of imprecise, marginal syllables for oral articulation.

Several recent studies have discussed the potential relationship between high rates of marginal syllables with later speech motor involvement in infants at risk for CP as a potential precursor to dysarthria (Long & Hustad, 2023; Ward et al., 2023). Additional prospective work in this area is necessary to explore the predictive value of Highest Ratios or Established Levels on the degree of speech motor involvement in children with CP. Specifically, do children with CP with little to no speech motor involvement produce more developmentally complex vocalizations in infancy than those with more moderate and severe levels of speech motor involvement? Also, is the frequency of advanced vocal forms (Highest Ratios) any more predictive than measures of vocal stage attainment (Established Levels) of speech motor impairment in children with CP? What is the time frame in which low rates of complex vocal

forms move from being a delay to something pathological, and how do we monitor this clinically? Notably, none of the children in our sample had a Highest Ratio or Established Level at either Level 4 or Level 5, highlighting the importance of examining the prognostic value of these variables across all stages of vocal development against impairment severity.

Regarding consonant diversity, 72% of our sample produced no utterances with consonants, and only two children produced more than one different true consonant. This reflects the low frequency of vocalizations at higher stages of vocal development involving canonical syllables. We similarly interpreted these results as a reduced ability to execute rapid and precise articulatory movements to produce a range of different consonants. This is in line with prior work showing reduced articulatory precision in children with CP who have speech motor impairments (Allison & Hustad, 2018a, 2018b; Hustad et al., 2020; Mahr et al., 2020). Additionally, the limited oral articulatory control observed among children in this study may be associated with an inability to produce lingual and labial movements independent of the jaw. This could explain the high proportion of bilabials in the small group of children at Level 3 and the large number of children at Levels 1 and 2. However, research is needed to further examine this possibility. Notably, previous studies excluded children with anarthria because the tasks of interest necessitated some functional speech. Using a vocal measure developed for use with prelinguistic infants therefore provides a novel approach to explore articulatory functioning and consonant diversity in non-speaking children. Ongoing work in this area may examine the predictive value of early consonant inventories on later speech motor involvement in children with CP. Future work on vocal predictors of anarthria may also focus on characterizing vowel and vowel-like features of utterances at Levels 1 and 2.

Our study also explored volubility, with findings indicating variable vocal rates among children in our sample. On average, children in our sample produced 5 utterances per minute. Relatively low vocal rates of four-year-old children with CP and anarthria may reflect broad neurological impact on speech motor control across the respiratory and phonatory subsystems. Additional factors such as positioning, postural control, and neck and trunk stabilization add an additional layer of complexity to quantifying vocal rates within a sample of children that also demonstrated significantly reduced gross motor functioning (>60% were classified as GMFCS Levels IV-V). As a result, the utility of vocal rate as an appropriate or useful measure is uncertain. Future work examining the predictive value of vocal rates for speech motor functioning in children with CP may seek to systematically manage the physical positioning of children.

While we found clear vocal characteristic trends across our sample, it is important to note that this group of children remains highly heterogenous. The variable volubility of our sample suggests a wide spectrum of phonatory and respiratory functioning, while vocal complexity and consonant diversity also indicates somewhat variable articulatory functioning. Also, highly heterogenous profiles of intellectual and language abilities are known to occur across children

with CP and anarthria (Fluss & Lidzba, 2020; Molinaro et al., 2020). Language and cognitive abilities can affect auditory perception, phonological awareness, and speech sound production (Peeters et al., 2008). Our sample predominantly had children with low language comprehension abilities, therefore the impact of language and cognition cannot be ruled out as potential causal variables associated with the findings of this study. Specifically, early vocal development is difficult to separate from early language development as the end goal is the production of words that will enable interaction. Accurately assessing cognitive and language abilities remains challenging for children with motor impairments, particularly for those who are unable to speak. Validated spoken language assessments for children with limited mobility now exist (Bootsma et al., 2023; Geytenbeek et al., 2010, 2014), but much work remains in developing reliable methods for assessing intellectual and language abilities in children with low motor functioning to support participation and improve long-term outcomes (Stadskleiv, 2020).

A secondary finding from our study was that three-quarters of our sample (74%) had a history of seizures. Previous research has indicated a deleterious impact of seizures on speech and language development in children with various neurodevelopmental disabilities (Allison et al., 2023; Coleman et al., 2013; Hidecker et al., 2018; Vaillant et al., 2022; Zhang et al., 2015). However, the relationship between seizures and anarthria has not been explicitly studied in children with CP to our knowledge. The high prevalence of seizures in our sample highlights the importance of further investigation into this relationship, considering factors such as the type and timing of seizures, in conjunction with other risk factors and co-diagnoses of CP. Understanding potential determinants of anarthria is crucial for identifying children who are likely to benefit from earlier referrals to speech therapy and interventions to improve functional communication through the use of AAC.

Few children in our sample were receiving AAC services (36%) despite a much larger majority participating in speech therapy (90%). This suggests the ongoing prevalence of a wait-and-see approach to speech development before introducing AAC to children, which has the potential to negatively impact later outcomes (McIntyre et al., 2011; Smith & Hustad, 2015). It is crucial to recognize that examining these characteristics at four years of age is already beyond the time frame of early intervention in the US. Employing similar methods of perceptual vocal analysis in younger children, both retrospectively and prospectively, holds promise to support the integration of AAC into early intervention services. Future research may work to develop and validate assessment tools to capture emergent articulatory characteristics in vocal functioning aligned with the five stages of vocal complexity described in the SAEVD-R to enable clinical monitoring of early vocal and speech development in children at high risk for CP and speech motor impairments. Related classification systems of speech and communication functioning have been developed and are commonly used by medical specialists to describe the various levels of functioning across children with CP, although none yet have the potential to track emergent speech abilities beginning in infancy (Barty et al., 2016; Hidecker et al., 2011; Pennington et al., 2010).

Limitations and Future Directions

Our study had several limitations. We did not examine differences in intellectual, language, or other cognitive abilities among children and focused our attention on measures of vocal production related to speech motor functioning. It is widely recognized that children with CP can present with heterogenous profiles of intellectual and language abilities (Berninger & Gans, 1986; Molinaro et al., 2020; Sabbadini et al., 2001). Only three children in our sample had language comprehension age equivalencies consistent with their chronological age, which limited our capacity to explore vocal production differences of children by language ability. Also, we were unable to assess intellectual abilities in these children due to their low motor functioning. Future research could examine vocal differences across children with anarthria based on their language and intellectual abilities to work toward identifying specific communication profiles of children with anarthria.

We acknowledge that the measures and coding procedures were developed for research on prelinguistic infants, although several studies have applied these methods to study vocal characteristics of children at speaking ages (Ertmer et al., 2007; Ha, 2019; Hitczenko et al., 2023; Lang et al., 2009; Morris, 2010). We intentionally selected these methods to facilitate a developmental assessment of vocal characteristics in non-speaking children. Nevertheless, developmental differences in respiratory, phonatory, and articulatory control may exist between infants and older children, regardless of the level of neurological involvement impacting speech subsystems. Recent research has indeed revealed ongoing maturation of speech development throughout childhood, even in children with severe speech motor impairments (Long, Mahr, et al., 2022; Mahr et al., 2020). Despite these potential differences, our findings align with previous trends of vocal developmental delays in children with or at risk for CP and speech motor impairments, highlighting the utility of these measures in future prospective work on this topic (Long, Christensen, et al., 2023).

Conclusion

Children with CP and anarthria tend to produce vocalizations representative of the two earliest stages of infant vocal development, demonstrating limited articulatory development in early childhood. Additional prospective work aiming to identify specific vocal markers for anarthria in infants at risk is necessary. Children with or at risk for CP demonstrating substantially reduced vocal developmental complexity such as limited consonantal features in vocal production well beyond expected ages of emergence are appropriate candidates for the early integration of AAC in therapies to enhance communication outcomes.

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Data Availability Statement

Raw audio recordings are identifiable and restrictions on access are required to protect the privacy and confidentiality of participants due to HIPAA requirements. Materials used to analyze these data are available at doi.org/10.17605/osf.io/7r5uk.

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