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Marginal and Canonical Babbling in 10 Infants at Risk for Cerebral Palsy

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ABSTRACT

Purpose: This study is a preliminary quantification and characterization of the development of marginal and canonical syllable patterns in 10 infants at risk for cerebral palsy (CP).

Method: We calculated marginal and canonical babbling ratios from parent–infant laboratory recordings of 10 infants at two time points, approximately 13 and 16 months of age. The frequency and diversity of labial, coronal, and velar types of marginal and canonical syllables were also examined. Differences across three outcome groups were compared: infants later diagnosed with CP (n = 3, CP group), risk of CP due to ongoing gross motor delays (n = 4, risk group), and current typically developing status with resolved gross motor delays (n = 3, TDx group). Performance on the Mullen Scales was included for perspective on cognitive development.

Results: Higher marginal syllable ratios were observed in the CP and risk groups than the TDx group. An increasing canonical syllable ratio across the two ages was consistently observed in the TDx group. The TDx group produced a greater frequency and diversity of canonical syllable types than the risk and CP groups, and of marginal syllable types than the CP group.

Conclusions: This study offers preliminary support for the possibility that speech motor impairment in infants with CP have the potential to be observed and quantified early in vocal development prior to the expected onset of first words. Prolonged rates of marginal syllable forms may be suggestive of speech motor impairment; however, additional longitudinal outcome data over a longer time course and a larger sample of infants are needed to provide further support for this possibility.

Between 60% and 90% of children with cerebral palsy (CP) have comorbid speech impairments that can affect communication effectiveness, educational participation, and social interaction throughout the life span (Mei et al., 2014, 2015; Parkes et al., 2010; Sigurdardottir & Vik, 2011). Speech and language impairments can be identified in children with CP as young as 2 years of age (Hustad et al., 2014, 2017). Such impairments prior to age two can be difficult to identify due to many factors, including wide ranging variability among children in early patterns of development, developmental delays due to prematurity, and variability in the severity and types of CP (Hustad et al., 2015; J. Lee et al., 2014). The emergence of articulatory abilities in infancy is of particular interest for the early identification of communication disorders in CP. Specifically, the study of infant vocal development may offer insight into the developmental trajectories of speech—from mild speech motor impairment to anarthria—even prior to the onset of the first word. At present, few studies have quantitatively examined vocal developmental patterns in this population under 24 months of age (Levin, 1999; Otapowicz et al., 2005). A prospective longitudinal investigation of vocal development in infants at risk for CP may offer important insights that advance early identification of speech impairment and support diagnostic and treatment planning for speech and augmentative and alternative communication at the earliest age possible.

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Perceptual Study of Prelinguistic Vocal Development: Articulation Domain

The study of prelinguistic vocal behaviors in clinical populations is theoretically informed by the longstanding body of research in typical infant vocal development. Stages of prelinguistic vocal development are well known to emerge across the first year of life prior to the onset of first words around 12 months (Koopmans-van Beinum & van der Stelt, 1986; Oller, 1978, 2000; Stark, 1980). The emergence of increasingly advanced vocal stages across the first year is theorized to evidence gradual control over the respiratory, phonatory, articulatory, and intonational domains to support mature speech (Oller, 2000). Although all speech subsystems have the potential to be impacted in CP due to neurological damage, we are particularly interested in the development of articulatory capabilities in infant babbling as a preliminary assessment of perceptual biomarkers of speech motor impairment. In part, this is because the articulatory subsystem has consistently been identified as making the greatest contribution to intelligibility reductions in preschool and school aged children with CP (Allison & Hustad, 2018; Chen et al., 2018; J. Lee et al., 2014).

In the articulatory domain in typical development, infants begin "cooing" between 1 and 4 months of life with limited, primitive articulatory vocal movements with the lips and tongue blade during phonation. The term *cooing* comes from the salient sound that occurs when the dorsum of infants' tongue makes contact with the posterior oral cavity during phonation resulting in a "coo" or "goo" sound. Between 3 and 8 months, infants transition into the expansion stage of vocal development. During this stage, infants practice and play with sounds such as raspberries, trills, clicks, and with articulatory and phonatory characteristics of speechlike sounds, including amplitude (yells and whispers), pitch (squeals and growls), and eventually *marginal babbling*. Marginal babbling in the present paper is defined as the production of slowly articulated primitive syllables, that is, syllable forms with a slow transition (perceived as > 250 ms) between the consonant-like element (i.e., margin) and vowel (Oller, 1980, 2000). Beyond this stage, the onset of *canonical babbling*—typically between 7 and 10 months of age—represents the emergent capacity to produce sounds that resemble mature speech. Canonical babbling is defined as the production of syllables with a fully resonant vowel and rapid transition (< 250 ms) between the consonant and vowel, for example, [ba] or [dada]. The timely onset of canonical babbling between its expected ages of emergence is well established to be a robust predictor of typical speech development (Morgan & Wren, 2018; Nathani et al., 2006; Oller & Eilers, 1988). Delays in the emergence of canonical babbling are frequently associated with later detected speech and language disorders (Lang et al., 2019; Nyman & Lohmander, 2018; Oller et al., 1998; Overby et al., 2019).

The transition through these vocal stages are considered infrastructural, that is, foundational to the later emergence of word production and mature speech (Oller et al., 2016; Stark, 1981). This study is based in the assumption that early neurological damage to the speech subsystems has the potential to deleteriously affect the trajectory of vocal development to support functional speech. Infants who experienced pre- or perinatal brain injury affecting motor development (as in the case of CP) are at an increased risk for later dysarthria and other motor speech disorders. However, few studies have empirically examined infant vocal development at any stage of development in CP.

Previous Research on Vocal Development in CP

On the study of cooing, Otapowicz et al. (2005) observed delayed cooing in 50% of children with CP through retrospective chart review and 96% of those with delays were later diagnosed with dysarthria. However, "cooing" in this study was not well defined and likely referred to babbling more broadly. In a seminal paper on babbling in CP, Levin (1999) cross-sectionally examined volubility and vocalization ratios produced by eight 12-month-old infants with CP. These infants produced more quasi-resonant vowels (i.e., phonation with vocal tract closure) than fully resonant vowels (i.e., phonation with an open vocal tract) and more marginal syllables than canonical syllables. Only two infants had reached the canonical babbling stage by 12 months—as measured by the 0.20 criterion ratio calculated as the number of canonical syllables/total number of utterances (Molemans et al., 2012; Oller & Eilers, 1988)—suggesting delays in its emergence in the majority of this sample. These infants produced single canonical syllables using a limited phonetic repertoire (predominately velar and labial sounds) compared with typically developing expectations of multisyllabic, variegated babbling with labial, alveolar, and nasal stops and nasal consonants by 12 months (Morgan & Wren, 2018).

Several other studies examining infants with neurodevelopmental delay (including CP) have also observed consistent canonical babbling delays and restricted phonetic repertoires compared with typical infants (Lohmander et al., 2017; Nyman & Lohmander, 2018). Only one study has shown a comparable age of onset for reduplicated canonical babbling between preterm infants with and without CP (approximately 7 months, per parent report of milestones), although later language milestones were delayed in the CP group compared with the no-CP group (Largo et al., 1986). We know of only one study that has investigated the longitudinal emergence of vocal behaviors in CP. Using the Infant Monitor of vocal Production (IMP), a recently developed interview-based instrument, Ward et al. (2022) observed parent-reported delays in the emergence of early vocal behaviors at 9 and 12 months in 18 infants at risk for CP compared with controls.

Despite this body of research, there is presently no published research on the longitudinal quantification of marginal and canonical babbling in CP. The present paper aims to examine articulatory characteristics of both marginal and canonical babbling in infants prospectively identified as at risk for CP at two time points in late infancy. The study of phonetic and acoustic characteristics of babbling have been previously reviewed in depth, offering fundamental insight into the kinematics of infants' oral movements (Rvachew & Alhaidary, 2018; Rvachew & Brosseau-Lapré, 2016). The present article, however, uses a perceptual approach to examine the emergence of articulatory control in CP. Specifically, we operate under the assumption that human listeners are uniquely attuned to the global features of syllable categories to support word learning and to monitor developmental progress as potential caregivers (Long et al., 2019; Ramsdell et al., 2012). Thus, naturalistic listening of infant babbling more closely matches that of parental report on infants' vocal developmental patterns for clinical screening. We recognize that there are limitations to the use of perceptual methods in the analysis of prelinguistic productions. However, given the powerful impact that clinically accessible perceptual methods could have on early identification, we sought to determine whether perceptual evidence for, or precursors to, speech motor impairment might be identifiable before the emergence of first words.

In this study, we examined marginal and canonical babbling development across two time points in a group of infants prospectively identified as being at risk for CP. Because these data were selected from a larger longitudinal study, we classified infants into three groups based on their most recent CP status: those with a confirmed CP diagnosis (CP group), those with an ongoing risk for a CP diagnosis (risk group), and those whose motor delays have resolved despite an early risk status for CP (TDx group). We hypothesized that infants in each group would reveal marginal and canonical babbling patterns predictive of their later clinical status. Specifically, we anticipated lower marginal and canonical babbling ratios in the CP group and higher ratios in the TDx group, in line with previous research indicating greater delays in later detected disorders. We also hypothesized that the CP group would produce the lowest frequency and diversity of sounds across the syllable types observed at both time points, whereas the TDx group would produce the highest overall frequency and diversity of syllable types, based on previous findings indicating reduced phonetic repertoires in CP around 12 months (Levin, 1999). Our research questions were as follows:

1. What are the patterns of marginal and canonical babbling change over time?
2. What are the frequency and diversity patterns of three primitive marginal syllable types (labial, coronal, and velar) of infants across groups?
3. What are the frequency and diversity patterns of three well-formed canonical syllable types (labial, coronal, and velar) of infants across groups?

Method

This study was approved by the institutional review board at the University of Wisconsin–Madison. Informed consent was received from all families prior to participation.

Participants

The participants were 10 infants (five male (M), five female (F)) under 24 months of age. These infants were selected from a larger, longitudinal speech project for inclusion in this study. Infants were recruited through local and regional medical centers in the midwestern region of the United States to participate in an ongoing longitudinal project (N = 51) on the study of the emergence and acquisition of speech in infants at risk for CP between 0–5 years of age. CP risk status was based on medical records and birth history (further expanded for each infant below). All infants were from homes where American English was the primary language. Eight infants were White, one infant was White and Pacific Islander, and one infant was White and Hispanic. All infants were born in the United States between 2016 and 2018.

During participation in the longitudinal speech project, caregivers brought their infant to the laboratory every 1–2 months to participate in a variety of formal and informal tasks including an unstructured, parent–infant (PI) interaction session. Our sample of 10 infants were selected from the larger cohort for having completed two PI interaction sessions at gestational age adjusted time points between 11 and 15 months (hereafter, "Time 1") and between 15 and 17 months (hereafter, "Time 2"). All 10 infants contributed one visit at each of the two time points observed, summing to 20 total visits (see Table 1). Professional-quality digital audio and video equipment was used to collect recordings of each PI interaction. During these sessions, caregivers were instructed to engage with their infant using toys, books, or simple games. Caregivers were not aware of the goals of this study and only

Table 1. Ages of infants at each time point observed.

Group	Infant	Age (months)	
		Time 1	Time 2
CP	1	13	16
	2	13	16
	3	12	16
Risk	1	12	15
	2	14	17
	3	14	16
	4	14	16
TDx	1	14	16
	2	11	16
	3	15	17

Note. CP = cerebral palsy; TDx = typically developing status.

instructed to engage with their infant naturalistically. The average length of recording analyzed for this study was 12 min (*SD* = 2.6).

Outcome Groups

Although data were prospectively collected, we classified infants into three groups based on their most recent diagnostic or risk status for group-level comparison from parent-reported and medical information collected in the longitudinal speech project. All infants were reported to have hearing within normal limits per newborn hearing screening tests and no notable hearing concerns reported by the parent. Infants' birth and medical history were described by parents during each laboratory visit intake. Relevant case history information was summarized for each participant based on this information and in consultation with the research speech-language pathologist familiar with each child across respective outcome groups below. Gross Motor Function Classification System Expanded and Revised (GMFCS-E & R; Palisano et al., 2007) levels were based on a consensus rating from parent and research SLP judgments using the "Before 2nd birthday" age band descriptors.

Three infants (two M, one F) were classified into the "CP group" because they had received a definitive CP diagnosis by the time this study was initiated. Four infants (F) were classified into the "risk group" because they had not received a definitive CP diagnosis but were still being monitored for ongoing gross motor delays and concern for CP. Three infants (M) were classified into the "TDx group" because they had caught up to typical gross motor developmental milestone expectations. The judgment for typical gross motor status was made based on the gross motor subtest on the Mullen Scales of Early Learning in conjunction with parent-reported information that the infant was no longer being followed for CP concerns. The three TDx infants had also been dismissed from participation in the longitudinal speech project because of their gross motor abilities. The label "TDx" is used to indicate that these infants were not prospectively recruited based on a typically developing status (rather risk for CP).

CP Group

Three infants comprised the CP group. CP-1 (M) was born full-term with no known complications. He was diagnosed with spastic CP at 5 months. He also had hypoxic ischemic encephalopathy (HIE), seizures, general hypotonia, oropharyngeal dysphagia, and reflux. He was fed through a gastrostomy tube from 6 weeks of age. At 46 months, his GMFCS-E & R was Level III; he was considered anarthric. CP-2 (M) was born 2 months prematurely and had a diagnosis of developmental delay and mild CP. Throughout infancy, he presented with low tone and mild hyponasality. At 47 months, his GMFCS-E & R was Level I; he was using 3–4 word sentences with some dysarthric and apraxic characteristics informally observed, although no formal diagnosis of either had been given. CP-3 (F) was born full term and experienced a perinatal stroke. She was diagnosed with right-sided spastic hemiplegic CP around 9 months. Throughout her participation in the longitudinal project, she presented with frequent drooling and a wet vocal quality. At 36 months, her GMFCS-E & R was Level II; she was producing some consonant–vowel–consonant (CVC) protowords.

Risk Group

Four infants comprised the risk group. Risk-1 (F) was born prematurely at 34 weeks as a twin birth. In infancy, she presented with mild general hypotonia and frequently demonstrated a wet vocal quality. By 42 months, her gross motor skills were within normal limits but continued to present with severely impaired articulation. Risk-2 (F) was born prematurely at 30 weeks and presented with flaccid tone throughout infancy. She was diagnosed with a rare genetic mutation and had secondary diagnoses of microphthalmia with myopia and nystagmus, oropharyngeal dysphagia, and global developmental delay. At 19 months, her gross motor skills were within normal limits, and she was not yet using any words although she was reportedly beginning to use reduplicated strings of [baba] canonical syllables. Risk-3 (F) was born at full term and had mild HIE at birth but largely demonstrated typical development throughout infancy. By 43 months, her gross motor skills were within normal limits, and she demonstrated age-appropriate articulation abilities. Risk-4 (F) was born full-term with heart decelerations during labor. By 29 months, she continued to present with mild gross motor delays. She was diagnosed with Angelman syndrome around 43 months and was anarthric.

TDx Group

Three infants comprised the TDx group. TDx-1 (M) was born full-term with abnormal positioning during delivery. Around 10 months, he presented with gross motor delays, mild right-sided weakness, and reduced proprioception. At 31 months, his gross motor skills were within normal limits, and he had with no overt oral motor or other speech concerns. TDx-2 (M) was born prematurely at 32 weeks as a twin birth. He presented with delayed crawling and weight shifting, spasticity, and reduced gross motor range of motion in early infancy. Around 18 months, his gross motor skills were within normal limits, with no overt concerns for speech development. TDx-3 (M) was born 2 months prematurely as a twin birth and early gross motor delays in infancy. Around 31 months, his gross motor skills were within normal limits, with no overt speech or language concerns.

Cognitive Assessment

To offer insight into potential cognitive influences on babbling in our sample, infants' performance on the Mullen Scales of Early Learning (Mullen, 1995) is reported in Table 2. The Mullen is a well-validated, standardized measure normed for infants and toddlers between 0 and 68 months of age across five areas of development: gross motor, fine motor, visual reception, expressive language, and receptive language. A T-score is calculated for each of the individual subtests and a total Early Learning Composite standard score is calculated from performance across all five subtests as a comprehensive measure of cognitive developmental ability. The average age of Mullen administration for the CP group was 13.2 months (*SD* = 1.2), for the risk group was 14.5 months (0.6), and for the TDx group was 12.3 months (1.5).

Vocal Coding Procedure

PI interaction sessions were extracted from the full laboratory recording for analysis. A primary (first author) and second reliability coder watched each extracted PI interaction to identify and label infant syllables in real-time for vocal characteristics within *The Observer XT* behavioral coding software (Noldus et al., 2000). The primary coder had extensive experience in syllable-level vocal coding according to the protocols described and used by Oller and colleagues from the Origin of Language Laboratory (Buder et al., 2013; Long, 2020; Nathani & Oller, 2001). These protocols were adapted for the purposes of this study to differentiate noncanonical and canonical syllables across vowel types and articulatory characteristics, further described below. The reliability coder was an undergraduate research assistant trained by the primary coder on the adapted protocol with instructional video examples and regular feedback on practice training material. During formal coding of this study, both coders were blinded to the age and outcome groups of infants although video information can influence observers' judgment about this information.

Eleven vocal types were used to categorize syllables in real-time during video observation based on the principles and properties of infant vocal development as defined by Oller (2000), described in Table 3. These included two vowel types (quasivowels and full vowels), marginal babbling (MB; three primitive syllable types: labial, coronal, and velar), canonical babbling (CB; three well-formed syllable types: labial coronal, and velar), and three verbal types (verbal-words, verbal-phrases, and verbal-sentences). All MB syllables were defined as syllable forms with a slow transition (perceived as > 250 ms) between the consonant-like element and vowel (Oller, 1980, 2000). CB syllables were defined as a well-formed, adultlike articulation of a

Table 2. Mullen scores of 10 infants at risk for cerebral palsy (CP).

Group	Infant	Gross motor	Fine motor	Visual reception	Receptive language	Expressive language	Early learning composite
CP	1	20*	33*	30*	21*	31*	61*
	2	23*	38*	33*	35*	39*	84*
	3	20*	44	34*	40	21*	71*
Risk	1	23*	48	57	39*	43	94
	2	20*	22*	20*	34*	28*	57*
	3	28*	54	49	44	39*	93
	4	20*	54	20*	31*	28*	69*
TDx	1	48	65	57	40	66	114
	2	44	51	48	44	29*	86
	3	51	54	40	49	34*	89

Note. All subtests are reported as T-scores. Early learning composite scores are calculated as standard scores. Scores with asterisks (*) indicate below average (T-score: 40 = 1 *SD* below mean of 50; standard score: 85 = 1 *SD* below mean of 100). TDx = typically developing status.

Table 3. Eleven syllable and verbal types used for infant vocal coding.

Syllable type	Definition
Quasivowel	Vowel-like sounds with a quasi-resonant nucleus with muffled resonance, often from a closed vocal tract at rest
Full vowel	Vowel sound with a fully resonant nucleus produced with an open vocal tract
MB-labial	Consonant-like element with slow formant transition to vocalic nucleus (partial or full bilabial contact)
MB-coronal	Consonant-like element with slow formant transition to vocalic nucleus (partial or full contact with front tongue and alveolar ridge/hard palate)
MB-velar	Consonant-like element with slow formant transition to vocalic nucleus (partial or full contact with back tongue and velum)
CB-labial	Consonantal element with adultlike formant transition to vocalic nucleus (full bilabial contact) (e.g., [ba] or [ma])
CB-coronal	Consonantal element with adultlike formant transition to vocalic nucleus (full contact with front tongue and alveolar ridge/hard palate) (e.g., [ta], [da], or [na])
CB-velar	Consonantal element with adultlike formant transition to vocalic nucleus (full contact with back tongue and velum) (e.g., [ka] or [ga])
Verbal-word	Single word approximation (e.g., “ball”)
Verbal-phrase	Two-word approximation (e.g., “want ball”)
Verbal-sentence	Three or more-word approximation resembling complete sentence (e.g., “me want ball”)

Note. MB = marginal babble; CB = canonical babble.

CV syllable with a quick formant transition (often perceived as < 250 ms) between the consonant and vowel. Note that although prelinguistic vocal coding was completed at the syllable level, linguistic verbal productions (words, phrases, sentences) were labeled at the utterance level. Phonatory characteristics such as pitch (e.g., growls, vocants, and squeals) were not differentiated in this study. Vegetative sounds (e.g., coughs, hiccoughs), phonation during mouthing of objects, and other nonspeechlike vocalizations (e.g., isolated labial trills, effort grunts, ingresses) were excluded from the coding protocol.

Several studies have examined characteristics of consonants produced during canonical babbling using phonetic transcription (Davis & MacNeilage, 1995; Rvachew & Alhaidary, 2018; Willadsen et al., 2020). It is widely agreed across the field of infant vocal development that broad phonetic transcription is inappropriate for portraying primitive (i.e., marginal) vocalizations because it inappropriately generalizes articulatory parameters of mature speech onto prelinguistic utterances (Rvachew & Alhaidary, 2018; Rvachew & Brosseau-Lapré, 2016). For this reason, we have used broad articulatory placement types (i.e., labial, coronal, and velar) to characterize the frequency and diversity of marginal and canonical syllables produced by infants at risk for speech motor impairment.

Babbling Ratios

The two primary variables of interest for our first research question were marginal and canonical babbling. A *marginal babbling ratio* (MBR) and *canonical babbling ratio* (CBR) was calculated for each infant at each age. Each respective ratio was calculated as the total sum of all marginal or canonical syllables divided by the total number of all syllables produced within that recording. Part-to-whole ratios were selected as the most commonly used calculation of proportional occurrence of specific vocal types (C. C. Lee et al., 2018; Nyman & Lohmander, 2018), thus supporting cross-study comparisons. Although the three verbal types were coded at the utterance level, these codes were included in the calculations for MBR and CBR to represent the total proportion of all vocalizations produced. As will be discussed in the results, the extremely low number of occurrences of any verbal type is believed to have had a negligible effect on either ratio for any infant.

A CBR around 0.15 is the commonly accepted criterion corresponding to the level at which infants can be judged to have reached the onset of the canonical babbling stage (Nathani et al., 2006; Oller et al., 1998). This criterion has ranged from 0.14 to 0.20 in previous studies examining canonical babbling onset in developmental disorders, with higher criteria often using utterance count as the denominator (Molemans et al., 2012; Nyman et al., 2021; Rvachew et al., 2005). We selected 0.15 as the standard criterion for laboratory-based recordings with syllable count as both the numerator and denominator. A discrete criterion has not been empirically evaluated for MBR and was, therefore, not used to gauge onset of marginal babbling in this study. Previous research on the consolidation of canonical babbling has suggested that in typical development, MBRs are expected to decrease and CBRs to increase over time as infants learn to coordinate the vocal tract for the production of mature speech (Rvachew & Brosseau-Lapré, 2016).

Coding Reliability

We randomly selected 40% of recordings (n = 8) for reliability coding. We calculated the interrater reliability of the total vocal type counts across the 11 syllable and verbal types coded for these sessions with the intraclass correlation coefficient (Shrout & Fleiss, 1979). We used a single score, absolute agreement, two-way random effects model and found good reliability between the two raters, ICC (2, 1) = .869, 95% CI [.801, .914].

Analysis

The analyses of this study are descriptive due to the preliminary nature of the study, and the small number of infants examined. Our particular interest was the examination of individual infants and the extent to which their vocal development was similar to, or different from, the others. Specifically, to examine the patterns of marginal and canonical babbling change over time, we compared the trajectories of marginal and canonical babbling ratios between two time points in infants across the three groups. To examine the frequency and diversity patterns of marginal and canonical babbling syllable types, we compared changes in the total number of each type produced by infants at each time point.

Results

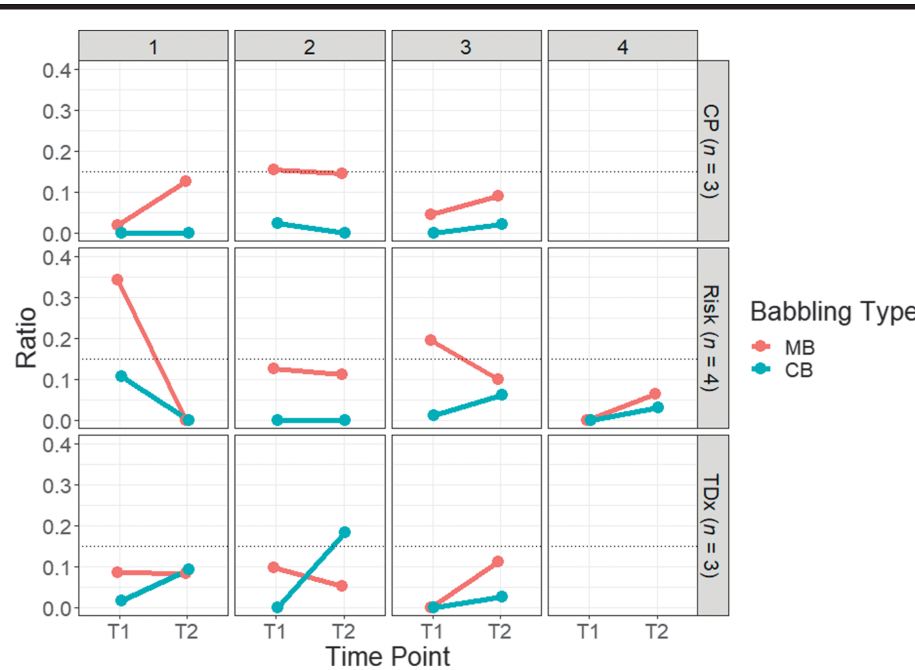
What Are the Patterns of Marginal and Canonical Babbling Change Over Time?

For the first research question, we sought to quantify the longitudinal development of marginal and canonical babbling in 10 infants prospectively identified as being at risk for CP. Figure 1 illustrates individual infants' MBR and CBR changes between two time points (approximately 13 and 16 months of age) that are grouped by outcome status. The dotted black line represents the 0.15 criterion for CBR.

All three infants in the CP group had higher MBRs than CBRs at both ages. For marginal babbling, 2/3 infants had a higher MBR at Time 2. Specifically, CP-1 and CP-3 increased their MBRs between the two time points (0.02 to 0.13 and 0.04 to 0.09, respectively), whereas the MBRs of CP-2 remained comparably high around 0.15 at both ages observed. These data indicate increasing or relatively stable MBRs across the two time points in the CP group. The CBRs of the three infants in the CP group were 0.02 or below at both time points. CP-1 had a CBR of 0.00 at both ages observed. CP-2 decreased their CBR from 0.02 to 0.00 and CP-3 increased their CBR from 0.00 to 0.02 between the two time points, indicating very low CBRs overall for all three infants at the ages observed.

In the risk group, 3/4 infants had a lower MBR at Time 2. Risk-1 had the highest MBR (0.34) at Time 1 out of the four infants that decreased to 0.00 by Time 2. Risk-3 had the second highest MBR (0.20) that then decreased to 0.10. Risk-2 also decreased their MBR from 0.13 at Time 1 to 0.11 at Time 2. Risk-4 had the lowest MBR at Time 1 that slightly increased to 0.06. Only Risk-3 and Risk-4 increased their CBRs from 0.01 to 0.06 and 0.00 to 0.03, respectively. The CBR of Risk-1 decreased from 0.11 to 0.00, and Risk-2 produced a CBR of 0.00 at both time points. These data indicate overall lower CBRs than MBRs of the risk group.

Figure 1. Marginal babbling (MB) ratios and canonical babbling (CB) ratios of infants across three outcome groups at two time points between 11 and 17 months. CP = cerebral palsy; TDx = typically developing status.



In the TDx group, 2/3 infants had a lower MBR at Time 2 than Time 1. TDx-1 had a relatively stable MBR around 0.08 at both time points; TDx-2 decreased from 0.10 to 0.05; and TDx-3 increased from 0.00 to 0.11. All MBRs of the TDx infants were over 0.05 except for one time point, (TDx-3, Time 1, MBR = 0.00). All three TDx infants increased their CBRs by Time 2 (TDx-1: 0.02 to 0.09; TDx-2: 0.00 to 0.18; TDx-3: 0.00 to 0.02). Out of all 10 participating infants, only TDx-1 and TDx-2 had a higher CBR than MBR at Time 2.

What Are the Frequency and Diversity Patterns of Three Primitive MB Syllable Types (Labial, Coronal, and Velar) of Infants Across Groups?

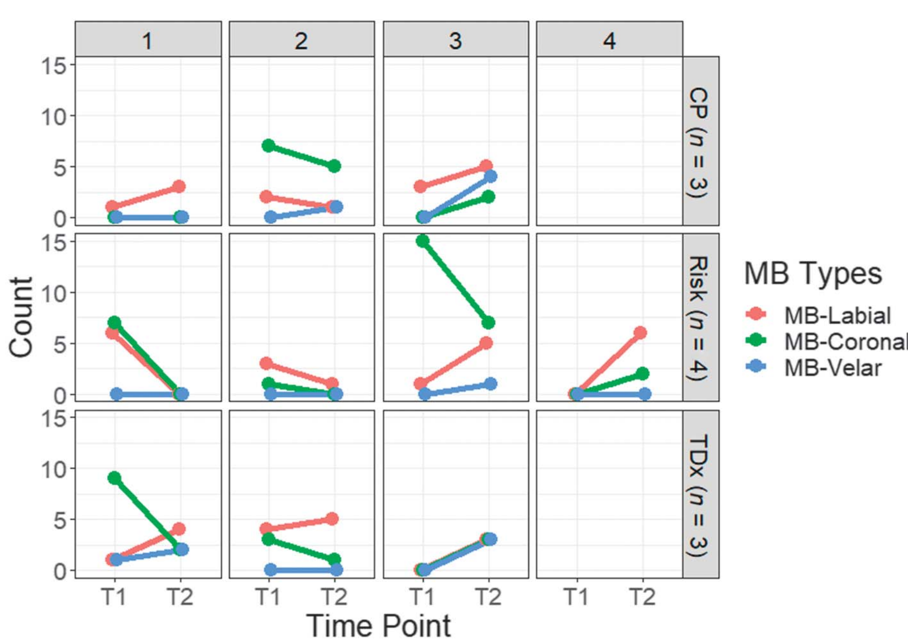
For the second and third research questions, we sought to characterize the frequency and diversity of marginal and canonical syllable types in infants across the two time points observed. We first calculated the total number of primitive syllable types (MB-labial, MB-coronal, MB-velar) produced by all infants (see Figure 2). Counts of each syllable type were used (instead of ratios) because of the overall low frequency of occurrence.

The CP group as a whole produced a total of 15 MB-labial types, 14 MB-coronal types, and five MB-velar types across both ages. CP-2 produced the highest total number of MB syllables (*n* = 16) followed by CP-3 (*n* = 14). CP-1 produced the fewest number of MB syllables (*n* = 4). CP-1 produced slightly more MB-labial syllables at Time 2 (MB-labial: one to three) and did not produce any MB-coronal or MB-velar syllables at either age observed. CP-2 produced fewer MB-labial and MB-coronal syllables at Time 2 (MB-labial: two to one; MB-coronal: seven to five) and produced one MB-velar syllable at Time 2 only. CP-3 produced more of all three syllable types at Time 2 (MB-labial: three to five; MB-coronal: zero to two; MB-velar: zero to four).

The risk group produced a total of 22 MB-labial types, 32 MB-coronal types, and one MB- velar type across both ages. Risk-3 produced the highest number of MB syllables (*n* = 29) followed by Risk-1 (*n* = 13), Risk-4 (*n* = 8), then Risk-2 (*n* = 5). Risk-1 produced fewer MB-labial and MB-coronal syllables at Time 2 than Time 1 (MB-labial: six to zero; MB-coronal: seven to zero) and did not produce any MB-velar syllables at either age. Risk-2 also did not produce any MB-velar syllables at either time point (MB-labial: three to one; MB-coronal: one to zero). Risk-3 produced more MB-labial and MB-velar syllables but fewer MB-coronal syllables at Time 2 (MB-labial: one to five; MB-coronal: 15 to seven; MB-velar: zero to one). Finally, Risk-4 produced more MB-labial and MB-coronal syllables at Time 2 (MB-labial: zero to six; MB-coronal: zero to two), but did not produce any MB-velar syllables at either age.

The TDx group produced a total of 17 MB-labial types, 18 MB-coronal types, and six MB-velar type across both ages. TDx-1 produced the highest number of MB

Figure 2. Total count of labial, coronal, and velar marginal babbling (MB) syllable types produced by 10 infants at two time points between 11 and 17 months. CP = cerebral palsy; TDx = typically developing status.



syllables (n = 19) followed by TDx-2 (n = 13). TDx-3 produced the fewest number of MB syllables (n = 9). TDx-1 produced more MB-labial and MB-velar syllables and fewer MB-coronal syllables at Time 2 than Time 1 (MB-labial: one to four; MB-coronal: nine to two; MB-velar: one to two). TDx-2 produced more MB-labial and fewer MB-coronal syllables at Time 2 (MB-labial: four to five; MB-coronal: three to one) and no MB-velar syllables at either age. TDx-3 did not produce any MB-labial, MB-coronal, or MB-velar syllables at Time 1 but produced three of each type at Time 2.

What Are the Frequency and Diversity Patterns of Three Well-Formed CB Syllable Types (Labial, Coronal, and Velar) of Infants Across Groups?

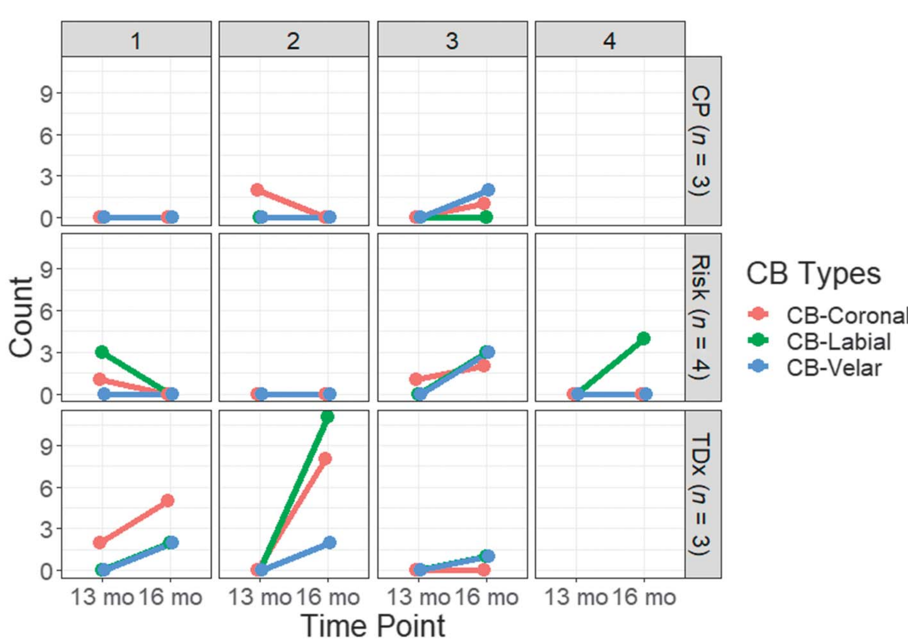
For canonical babbling, we calculated the total number of well-formed syllable types (CB-labial, CB-coronal, CB-velar) produced by infants at each time point to characterize the frequency and diversity of canonical syllable types (see Figure 3).

The CP group produced a total of zero CB-labial types, three CB-coronal types, and two CB-velar types across both time points. CP-3 produced the highest number of CB syllables (n = 3) followed by CP-2 (n = 2). CP-1 did not produce any CB syllables at either point. CP-2 produced two CB-coronal syllables at Time 1 but no other CB-syllable type at either point. CP-3 produced one CB-coronal syllable and two CB-velar syllables at Time 2, but no other CB-syllable type at either age observed.

The risk group produced a total of 10 CB-labial types, four CB-coronal types, and three CB-velar types across both time points. Risk-3 produced the highest number of CB syllable types (n = 9) followed by Risk-1 (n = 4) and Risk-4 (n = 4). Risk-1 produced fewer CB-labial and CB-coronal types at Time 2 than Time 1 (CB-labial: three to zero; CB-coronal: one to zero) and did not produce any CB-velar types at either point. Risk-2 (n = 5) did not produce any CB syllable types. Risk-3 produced more CB-labial, CB-coronal, and CB-velar types at Time 2 than Time 1 (CB-labial: zero to three; CB-coronal: one to two; CB-velar: one to three). Risk-4 produced four CB-labial types at Time 2, but no other syllable type at any other point observed.

The TDx group produced a total of 14 CB-labial types, 15 CB-coronal types, and five CB-velar type across both time points. TDx-2 produced the highest number of CB syllable types (n = 21) followed by TDx-1 (n = 1). TDx-3 produced the fewest number of CB syllable types (n = 2). TDx-1 produced more CB-labial, CB-coronal, and CB-velar types at Time 2 than Time 1 (CB-labial: zero to two; CB-coronal: two to zero; CB-velar: zero to two). TDx-2 also produced more of each type at Time 2 than Time 1 (CB-labial: zero to 11; CB-coronal: zero to eight; CB-velar: zero to two). TDx-3 produced one CB-labial and one CB-velar type at Time 2 and did not produce any other CB-syllable type at either time point observed.

Figure 3. Total count of labial, coronal, and velar canonical babbling (CB) syllable types produced by 10 infants at two time points between 11 and 17 months. CP = cerebral palsy; TDx = typically developing status.



Vowel and Verbal Types

To exemplify the proportional differences of vocalizations produced by infants in each group, vowel type ratios were also calculated (see Figure 4). All infants had higher quasivowel and full vowel ratios than MBRs or CBRs at both time points. CP-3 and Risk-4 slightly decreased their quasivowel ratio by Time 2, whereas Risk-1 slightly increased their full vowel ratio by Time 2. Overall, 6/10 infants had a higher quasivowel ratio and 7/10 infants had a lower full vowel ratio by Time 2. There were no remarkable patterns across the 10 infants observed in either vowel type ratio within or across groups.

The number of verbal types (words, phrases, and sentences) produced by our sample of infants was very low. Only two infants produced any single words (Risk-3: three words; TDx-2: six words) and one infant produced one two-word phrase (Risk-3). All linguistic utterances produced by these three infants occurred at Time 2 only.

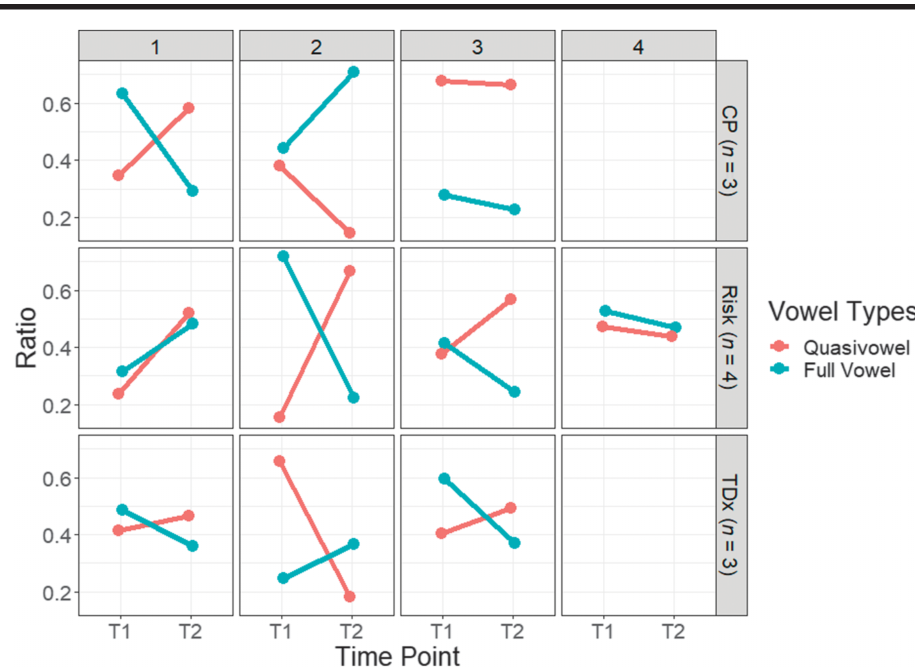
Discussion

This preliminary study is one of the first of its kind to examine marginal and canonical babbling at two time points in 10 infants at risk for CP during the second year of life using naturalistic listening methods. Specifically, we quantified the longitudinal development of marginal and canonical babbling and characterized the frequency and diversity of syllable types in 10 infants separated into three outcome groups across two time points between 11 and 17 months of age. We used a descriptive approach with a focus on individual infants because of the preliminary nature of this study. Overall, we found higher proportions of marginal babbling across both time points in the CP and risk groups, and a greater proportion of canonical syllables in the TDx group. Greater diversity of marginal syllable types was also observed overall in the CP and risk group, whereas the TDx group demonstrated somewhat more diversity of canonical syllable types, further discussed below. Individual infants' cognitive abilities across five areas of performance and overall production of vowel and verbal types offer perspective for interpretation.

Marginal and Canonical Babbling Ratios

We first examined the proportional change of marginal and canonical babbling across two time points in the three groups of infants. We hypothesized that the CP group would have lower MBRs and CBRs, and that the TDx group would have higher MBRs and CBRs at both time points. This hypothesis was based in the presumption that children with a CP diagnosis would have frank neurological impacts associated with their early diagnosis, which, in turn, would influence development—particularly speech motor development—relative to those who did not have a diagnosis. Although a certain amount of variability was observed, several patterns in line with our hypotheses

Figure 4. Quasivowel and full vowel ratios of 10 infants across three outcome groups between 11 and 17 months. CP = cerebral palsy; TDx = typically developing status.



were noted. In the CP group, we observed increasing or stable MBRs but very low CBRs ($\leq$ 0.02) across the two time points for all three infants. The risk group demonstrated decreasing MBRs in 3/4 of the infants and overall lower CBRs than MBRs. In the TDx group, 2/3 infants also showed decreasing MBRs but all three TDx infants had higher CBRs by Time 2. Also, the two infants in our sample with higher CBRs than MBRs at either time point observed were both TDx infants. We describe potential explanations subsequently.

The production of primitive forms of prelinguistic syllables is historically viewed as a required stage of development prior to the onset of more well-formed syllables (Koopmans-van Beinum & van der Stelt, 1986; Oller, 2000; Stark, 1980). Although marginal babbling has received limited investigation in clinical populations, several studies have shown that in later stages of infancy in typical development, marginal syllable rates should decrease as infants learn to coordinate the vocal mechanism throughout the stages of prelinguistic vocal development (Lynch et al., 1995; Nathani et al., 2006). This pattern was indeed observed in 2/3 TDx infants (whose motor delays eventually resolved); however, marginal babbling was largely predominant in the CP and risk group infants at both ages. Thus, our results support the notion that in infants whose early motor delays eventually resolve, the emergence of mature CV syllables is a robust phenomenon for typical speech development. These findings also highlight the potential role of neuroplasticity supporting developmental maturation in CP, particularly in the vocal domain (Hadders-Algra, 2014). Conversely, for infants with ongoing motoric delays, protracted high rates of marginal syllables throughout infancy may then suggest delayed or disordered speech development.

Frequency and Diversity Patterns of Marginal and Canonical Babbling Syllable Types

Several trends were also noted in the frequency and diversity patterns of the marginal and canonical babbling types produced by infants across the three groups. For marginal babbling, the risk group produced the highest number of MB-syllable types, followed by the CP and TDx groups, respectively. For canonical babbling, the TDx group produced the highest number of CB-syllable types followed by the risk and CP groups, respectively.

Within each group, individual differences were also observed. CP-1, who remained anarthric by 46 months produced the fewest MB-syllable types and no canonical syllables at any age. Risk-3, who produced the most MB- and CB-syllable types of the four infants in the risk group, was one of only two infants in our sample who produced at least one word. TDx-2, who produced the highest number of words out of the two linguistic infants, also produced the highest number of CB-syllable types across all 10 infants. It is worth noting that Risk-2 and Risk-4 demonstrated consistently lower MB- and CB-syllable types than the other two infants in the risk group. Both infants were the only two in our sample with co-morbid genetic conditions, which suggests a need for further study of the impact of genetic diagnoses and other comorbidities on vocal and speech development in CP.

The 10 infants in our sample produced more than two times as many MB syllable types (n = 130) as CB syllable types (n = 56). The gross overview of vowel types indicated higher quasivowel ratios than full vowel ratios, overall. Similar patterns (more marginal syllables and quasivowels than canonical syllables and full vowels, respectively) were also observed by Levin (1999), adding preliminary support to the idea that a reduced range of oral motor function may be evident in the production of more primitive forms of sounds by infants who receive an early CP diagnosis. A noted difference from our findings and the Levin (1999) study is the overall count of velar syllable types. Levin observed the highest frequency of velar sounds in her group of eight infants with a CP diagnosis. The MB- and CB-velar syllable types were the least frequently observed syllable type across all infants in our sample. Interestingly, the three TDx infants produced the highest number of both MB- and CB-velar syllable types, indicating greater overall diversity of syllable types in the TDx group than the CP or risk groups. These findings may be attributed to individual differences given the small sample sizes of both studies; however, additional study of the articulatory placement of consonant-like elements in syllables produced by infants at risk for CP is clearly warranted.

Results of this study may indicate that early features of what is later diagnosed as speech motor impairment have the potential to be observed in the babbling of infants at risk for CP. Specifically, we found that infants with greater risk (or a confirmed diagnosis) of CP generally produced more marginal syllables whereas the TDx group produced more canonical syllables. However, infants within each outcome group still showed wide variability in their use of both marginal and canonical syllables. These patterns may suggest that potential speech motor delay or impairment in the CP and risk groups may have impacted their ability to produce articulatory syllables with the precision necessary to be considered canonical. In other words, the protracted production of a high proportion of marginal syllables without expected progression to canonical syllables may be a reflection of speech motor involvement. This level of impairment may be expected to affect an infant's ability to produce rapid formant transitions in CV syllables, resulting in the perception of a greater proportion of marginally formed syllables. This finding would be consistent with literature demonstrating frequent articulatory impairment affecting vowels and consonants as a feature of dysarthria in individuals with CP (Mei et al., 2020; Nordberg et al., 2014; Platt et al., 1980). However, additional research involving a longer timeframe, encompassing the age of 4–5 years when dysarthria can be clinically diagnosed with certainty is necessary. In this study, we do not yet know which infants in our sample ultimately will be diagnosed with dysarthria and thus our observations must be considered speculative.

Cognitive Performance of Infants

It is important to emphasize that the two time points observed (encompassing 12–19 months overall) are beyond the expected ages of onset for both marginal and canonical syllable types, and only one of the infants in the TDx group reached the canonical babbling stage (using the 0.15 criterion) even by Time 2. Although Levin (1999) used a slightly different criterion, she similarly observed a delayed canonical babbling onset in only 2/8 of infants with CP. Furthermore, few infants in our sample produced any words during the recordings. To some extent, this may reflect the sampling environment associated with an unfamiliar laboratory setting, but infants' Mullen performance may also provide additional insight into this phenomenon. CP-1 and Risk-2, the two infants in each group with the lowest MBR and no CB syllables, both had the lowest Mullen Early Learning Composite scores in their respective groups. Conversely, CP-2 and Risk-3, the two infants in each group with the most stable or increasing MBRs and CBRs, demonstrated higher Mullen Early Learning Composite scores relative to other infants in their respective groups. Although all three TDx infants performed within an average range on the gross and fine motor subtests, 2/3 of the infants in the TDx Group demonstrated significantly below average expressive language abilities at the time of assessment (approximately 13 months), which resolved by the point of their dismissal from the larger longitudinal study; all three infants in the CP group and 3/4 of the risk group also demonstrated below average expressive language abilities.

Reduced expressive language performance in 8/10 infants in our sample may well be associated with speech motor impairment given that expressive language is demonstrated through speech production; however, data reported in this study do not allow us to differentiate between expressive language deficits and speech production deficits that mask the ability to express language. Ultimately, this is an important area for future research. It is noteworthy that previous research has indicated that children with conditions leading to an at-risk status for CP such as prematurity, perinatal stroke, and moderate–severe HIE often have expressive language challenges beyond infancy (Edmonds et al., 2021; Largo et al., 1986; Trauner et al., 2013).

Children with CP are heterogenous, and different constellations of speech and language profiles have been established in preschool children with CP (Hustad et al., 2010). At younger ages, it is difficult to distinguish between expected developmental variability and disorder for many children. In children and adults with CP in general, it can be difficult to adequately characterize underlying language and cognitive abilities in the presence of motor impairments that limit testing capabilities. However, 6/10 infants in our sample had higher receptive language than expressive language abilities. Interestingly, the TDx group infant who met the 0.15 criterion at Time 2 and who spoke approximately six words during their recording at Time 2 was one of these infants. Clearly, more research in prelinguistic vocal and early speech development of infants at risk for CP is needed to advance our ability to identify children with speech and language impairments at the earliest possible age to enable provision of early intervention services.

Limitations and Future Directions

Our findings provide preliminary support for our central hypothesis that precursors to later speech impairment in CP may be evident in prelinguistic vocal behaviors in infancy. Nonetheless, results should be interpreted with some caution because of the small sample size included. Also, we examined only two age points in infancy. Future studies should aim to study prelinguistic vocal development across a larger time frame in infancy to better understand biomarkers of speech impairment in this population.

A more fine-grained perceptual analysis of the predictive nature of specific canonical babbling patterns (e.g., reduplicated and variegated babbling) could also be studied in future work. Although this study used real-time, point-based coding, future studies should also compare syllable and utterance durations. Future studies could examine acoustic measurements of marginal and canonical syllables (e.g., timing of the second formant transition) for additional perspective. Examining these measures across multisyllabic utterances could potentially guide our understanding of the impact of CP on the full motor speech system, including respiratory support.

More clinically translatable perceptual tools than ratio calculations could also be used in future studies examining prelinguistic vocal development in infants at risk of CP or other clinical populations. Stark and colleagues have adopted a more motoric perspective in their study of infant vocalizations and developed the Stark Assessment of Early Vocal Development–Revised (SAEVD-R; Nathani et al., 2006) to categorize utterances according to the articulatory complexity of the sounds at the utterance level. Yet, few studies have utilized the SAEVD-R to measure infant vocalizations. Additional research using this or other prelinguistic classification tools could further specify supraglottal and phonatory characteristics of vocalizations in clinical populations, including CP. Finally, research has indicated that typical infants do not vocalize at comparable rates across laboratory and home settings (Lewedag et al., 1994; Oller et al., 2021). Thus, future research should also compare vocal behaviors of infants at risk for CP to TD controls across multiple environments.

Clinical Implications

The preliminary findings from this study may suggest that features of speech motor delay or impairment are evident even in the earliest vocal behaviors among infants with CP or those who are at risk for CP. Across our sample of 10 infants, we observed higher marginal babbling and lower canonical babbling in the CP and risk groups compared with the TDx group. Higher rates of marginal babbling in infants with ongoing risk or diagnosis of motor-based disorders like CP may indicate underlying neurological damage to the motor system affecting their production of well-formed, adultlike syllables. However, additional research is necessary to expand these descriptive observations in a larger sample. These findings indicate a critical need for the large-scale study of prelinguistic and early speech development in infants at risk for CP under 24 months.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

Acknowledgments

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