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Review Article

Vocal Characteristics of Infants at Risk for Speech Motor Involvement: A Scoping Review

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ABSTRACT

Purpose: The purpose of this scoping review was to (a) summarize methodological characteristics of studies examining vocal characteristics of infants at high risk for neurological speech motor involvement and (b) report the state of the high-quality evidence on vocal characteristic trends of infants diagnosed or at high risk for cerebral palsy (CP).
Method: The PRISMA (Preferred Reporting Items of Systematic Reviews and Meta-Analyses) extension for scoping reviews was followed for reporting our review. Studies measured prelinguistic vocal characteristics of infants under 24 months with birth risk or genetic conditions known to commonly present with speech motor involvement. Fifty-five studies met criteria for Part 1. Eleven studies met criteria for synthesis in Part 2.
Results: A smaller percentage of studies examined infants with or at risk for CP compared to studies examining genetic conditions such as Down syndrome. The median year of publication was 1999, with a median sample size of nine participants. Most studies were conducted in laboratory settings and used human coding of vocalizations produced during caregiver–child interactions. Substantial methodological differences were noted across all studies. A small number of high-quality studies of infants with or at risk for CP revealed high rates of marginal babbling, low rates of canonical babbling, and limited consonant diversity under 24 months. Mixed findings were noted across studies of general birth risk factors.
Conclusions: There is limited evidence available to support the early detection of speech motor involvement. Large methodological differences currently impact the ability to synthesize findings across studies. There is a critical need to conduct longitudinal research with larger sample sizes and advanced, modern technologies to detect vocal precursors of speech impairment to support the accurate diagnosis and prognosis of speech development in infants with CP and other clinical populations.

Infants at risk for cerebral palsy (CP) are also at high risk for speech motor disorders such as pediatric dysarthria, a condition characterized by imprecise articulation, slow rate, reduced intelligibility, speech sound distortions, as well as involvement of respiratory, phonatory, and resonatory subsystems for speech production (Allison & Hustad, 2018; McCauley & Strand, 2008; Mei et al., 2020; Odding et al., 2006). Neurological substrates of speech motor function impacted at birth are known to affect control over the oral motor system even when other cognitive and language processes are unaffected, which can have long-term implications for communicative functioning and participation across contexts (Haas et al., 2021; Schölderle et al., 2021). The early and accurate prediction of functional speech outcomes in children with CP remains a major challenge for speech-language pathologists because of the overlapping characteristics of speech motor impairment and typical development, alongside the

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adaptive plasticity of networks following neurological impact to the brain affecting developmental progress throughout infancy (Allison & Hustad, 2018; Haas et al., 2021; Johnston et al., 2009).

Recent research aiming to predict the level of speech motor involvement (i.e., restricted speech functioning across varying degrees of speech motor precision, stability, and control) in children with CP at the youngest possible age is promising. Specifically, speech intelligibility of children with CP at 2 years of age is highly predictive of their speech intelligibility at 8 years of age (Hustad et al., 2019, 2020; Mahr et al., 2020). Validated assessment tools such as the Profile of Childhood Apraxia of Speech and Dysarthria (Iuzzini-Seigel et al., 2022) and the Bogenhausen Dysarthria Scales–Childhood Dysarthria (Haas et al., 2020; Schölderle et al., 2020) were developed to characterize auditory-perceptual features of speech subsystem involvement for the differential diagnosis of pediatric speech motor disorders, yet these measures can only be used for children who are able to produce words. Children with CP are especially vulnerable to delays in speech referrals because of the clinical prioritization of gross and fine motor needs and greater number of targeted interventions to support these areas of development in children with CP under 2 years (C. Morgan et al., 2021). Consequently, early speech development in children with CP is often managed with a "wait-and-see" approach (McIntyre et al., 2011; Shevell et al., 2001; A. L. Smith & Hustad, 2015). Early and accurate detection of speech motor involvement before speaking ages could have an important impact on treatment and on long-term outcomes for children with CP.

A small body of evidence has emerged aiming to detect vocal precursors of speech motor involvement in infants under 24 months at risk for CP and other neurodevelopmental conditions. In a seminal paper on this topic, Levin (1999) observed low rates of canonical babbling (i.e., adult-like consonant–vowel syllables with rapid formant transitions) and monosyllabic consonant production in infants with CP at 12 months. Other recent studies have confirmed that comparing rates of infant canonical babbling can accurately differentiate children with neurodevelopmental disabilities (including CP) from typically developing children under 24 months of age (Lohmander et al., 2017; Nyman & Lohmander, 2018). Parent-reported onset ages of infant vocal milestones can also be highly predictive of speech outcomes across a range of children at risk for neurodevelopmental disabilities and speech motor involvement, particularly CP and Down syndrome (Locatelli et al., 2021; Lynch, Oller, Steffens, Levine, et al., 1995; Otapowicz et al., 2005).

Through this work, we have noted rising attention to the study of vocal characteristics of infants at risk for speech motor involvement. However, the state of the collective evidence is unknown because these studies have not been systematically reviewed. During a preliminary search of studies on this topic, we found a larger, longer-standing body of work examining vocal characteristics of infants with Down syndrome, a condition in which over 95% of children present with speech motor involvement (Wilson et al., 2019). Several other studies examined vocal characteristics of a broader group of children with "neurodevelopmental disabilities" including CP, which suggests a broader scope of study for mixed clinical groups of children with complex communication needs (Lohmander et al., 2017; Nyman & Lohmander, 2018). For these reasons, we set out to first review the methodological practices in a larger scope of studies across a broad range of clinical conditions at risk for speech motor involvement to understand the range of approaches used to study early vocal development in infants. This was necessary because prior reviews of other clinical populations (i.e., hearing loss, language differences, autism), typically developing infants, and infants across cultures have reported large methodological variability across studies (Bryant, 2022; Lang et al., 2019; McDaniel & Gifford, 2020; L. Morgan & Wren, 2018; Yankowitz et al., 2019).

In the present review, our primary interest was infants with CP. However, because CP is often not diagnosed in infants until 12–24 months in the United States (Novak et al., 2017; te Velde et al., 2019), we sought to summarize the research methodologies used to study vocal precursors to speech motor involvement. We then sought to evaluate the quality of the evidence that specifically addressed the narrower scope of infants diagnosed with CP. To meet this aim, our goals were twofold and separated into Parts 1 and 2, hereafter.

In Part 1, our first goal was to describe the broader populations of infants at risk for speech motor impairment and characterize research methods used to study vocal characteristics in infants at risk for speech motor involvement, including genetic and chromosomal conditions and birth risk conditions. We predicted that there would be a very small body of literature examining infants with CP. However, we expected to find a much larger body of literature in genetic and birth risk conditions that commonly present with speech motor involvement (i.e., Down syndrome) because these populations can be more easily identified and enrolled in research from birth. This larger scope allowed us to broadly synthesize the methodological landscape of the study of vocal precursors to speech motor involvement and identify methodological trends that may be applied to the study of vocal characteristics in CP.

In Part 2, our goal was to narrow our scope to the study of CP. Specifically, we sought to critically appraise and synthesize findings from high-quality studies identified

in Part 1 that examined infants diagnosed and at risk for CP to determine whether trends in delayed or unexpected vocal characteristics are evident compared to typically developing expectations across prelinguistic stages of development.

Key Questions

Part 1

1. What clinical populations known to present with neurodevelopmental speech motor involvement at later ages have been studied with respect to their early infant vocal characteristics?

2. What research methods have been used to study vocal characteristics across these populations?

Part 2

3. What is the state of the high-quality evidence on vocal characteristics of infants diagnosed or at risk for CP and speech motor involvement?

Part 1: Populations and Method

The PRISMA (Preferred Reporting Items of Systematic Reviews and Meta-Analyses) extension for scoping reviews was followed for reporting our review (Tricco et al., 2018). This method provided a systematic approach to identify studies examining vocal characteristics of infants at high risk for speech motor involvement (Part 1) and to synthesize findings across studies of infants diagnosed or at risk for CP (Part 2).

Method

Literature Search

The review team collaborated with a research librarian (second author) to develop and execute a comprehensive search of the literature. This search combined controlled vocabulary and title–abstract terms on the evaluation of infant vocal characteristics across broadly defined genetic and risk-based clinical populations commonly diagnosed with speech disorders. The following databases were searched from database inception through November 24, 2021: CINAHL Plus With Full-Text (EBSCO), Scopus (Elsevier), the Cochrane Central Register of Controlled Trials (Wiley), and Web of Science (Clarivate). On May 12, 2022, a revised search was conducted from database inception to include additional risk factors for CP. An age filter was applied to focus on infants younger than 2 years of age. No other publication type, language, or date filters were applied. Results were downloaded to a citation management software (EndNote) and underwent manual deduplication by the second author. Unique records were uploaded to the Covidence screening platform (Covidence Systematic Review Software, 2021) for independent review by the project team members. Title–abstract and full-text screening was conducted by the first author and two research assistants (RA1 and RA2) using this program using predetermined eligibility criteria described below.

Eligibility Criteria

Our primary eligibility criteria required that studies evaluate prelinguistic vocal characteristics of infants under 24 months of age in a clinical population with a diagnosis, or high risk of a diagnosis, associated with neurological speech motor involvement. The full eligibility criteria are summarized in Table 1 and described hereafter. The full list of search terms is provided in Appendix A.

Studies were eligible for inclusion if they characterized any aspect of infant vocalization as an independent or dependent variable, including canonical babbling ratios (CBRs), utterances per minute, or onset ages of vocal milestones. Our age criterion was initially set to include

Table 1. Eligibility criteria for Part 1.

Variables	Inclusion criteria	Exclusion criteria
Populations	Cerebral palsy, genetic and chromosomal conditions, preterm birth, and other studied developmental conditions associated with pediatric speech motor involvement and disorders	Conditions affecting language and social communication, not speech motor production (e.g., autism, fragile X syndrome, Rett syndrome), societal group comparisons (e.g., socioeconomic groups, poverty level), hearing loss, neurotypical development, and structural differences (e.g., craniofacial abnormalities, cleft palate)
Vocalization variable terms	Vocal development, articulation development, babbling, cooing, expansion stage, jargon, prelinguistic, preverbal, prespeech	Linguistic variables only (e.g., receptive/expressive language milestones, and linguistic measures of vocabulary, MLU, syntax, or pragmatics)
Age range	Prospective studies of human infants < 24 months of age; retrospective studies of vocal behavior occurring < 24 months	Only > 24 months of age

Note. Full search terms across databases are presented in Appendix A. MLU = mean length of utterance.

studies examining infants under 24 months; however, our search yielded several retrospective studies examining earlier vocal characteristics of older children with eligible clinical conditions. These studies were included if they reported prelinguistic vocal information at younger ages (e.g., caregiver-reported onset ages of vocal milestones in infancy).

During the title–abstract and full-text screening phases, studies examining diagnostic groups with a high prevalence of speech motor involvement (e.g., Down syndrome and CP) were eligible for inclusion. Common risk conditions for CP (e.g., preterm birth [< 37 weeks gestational age], very low birth weight [VLBW; < 1,500 g], brain lesions, hydrocephalus, failure to thrive, meningitis) were also eligible because of prior work establishing significant associations with CP (Alieva & Gasanova, 2015; Soleimani et al., 2014). Several populations emerged that were captured in our search but were ambiguous as to the extent of speech motor involvement (e.g., Cri-du-Chat syndrome, spina bifida, and congenital galactosemia). In these cases, project team members conducted an independent online search of research reporting speech disorders in that population through Google Scholar, PubMed Central, and information reported on that population listed on CDC.gov. If prior research indicated a presence of speech motor disorders in a specific population, that population was eligible for inclusion during the screening phases of our review. All final eligibility decisions were ultimately based on whether both screeners agreed that a study was eligible for inclusion across all relevant factors (population, vocalization variables, and ages studied). This iterative inclusionary method is supported by guidelines previously indicated for exploratory scoping reviews wherein the volume of the scope of certain criteria (e.g., relevant populations) is the empirical question and thus cannot be wholly defined a priori (Munn et al., 2018; Peters et al., 2020).

Reliability

Reliability was conducted on 100% of the articles at both the title–abstract and full-text screening levels. The Covidence software (2021) automatically calculates Cohen's kappa to measure the interrater reliability among pairs of coders. At the title–abstract screening level, the first author and RA1 (who each screened 96% of papers) had a Cohen's kappa of .37 (fair agreement). The first author and RA2 (who each screened 4% of papers) had a Cohen's kappa of .36 (fair agreement). At the full-text screening level, the first author and RA1 (who each screened 100% of papers at this level) had a Cohen's kappa of .75 (substantial agreement). The screeners met regularly to discuss ongoing discrepancies to prevent coder drift. Disagreements were settled through final consensus decisions among the pairs of screeners. The full output of reliability statistics calculated through the Covidence software is reported in the Supplementary Material.

Results

The PRISMA flow diagram for the inclusion of studies across these criteria is indicated in Figure 1. The database search yielded 8,908 records. Fifty-five articles met full criteria for inclusion. Data extraction occurred in the Covidence software platform.

Individual study characteristics are presented in Table 2. The median year of publication was 1999 (range: 1972–2022), and the median sample size was nine participants (range: 1–3,052) across the 55 studies.

Clinical Populations

The most common clinical populations studied were Down syndrome (n = 25), preterm birth (n = 13), and CP (n = 10).[1] The remaining studies examined vocal characteristics of other genetic, chromosomal, or birth risk populations. The average age ranges studied across groups were as follows: Down syndrome, 7–21 months; preterm birth, 6–16 months; CP, 11–26 months; other genetic/chromosomal disorders, 9–25 months; and other birth risk factors, 8–23 months. The genetic and chromosomal conditions previously studied were unspecified chromosomal syndrome (n = 3), Angelman syndrome (n = 1), sex chromosomal disorder (n = 1), Cri-du-Chat syndrome (n = 1), congenital galactosemia (n = 1), Prader-Willi syndrome (n = 1), and Smith-Magenis syndrome (n = 1). Birth risk conditions previously studied were brain injury (n = 3), failure to thrive (n = 3), VLBW (n = 3 papers), hydrocephalus (n = 2), microcephaly (n = 2), macrocephaly (n = 1), neonatal meningitis (n = 1), and spina bifida (n = 1).

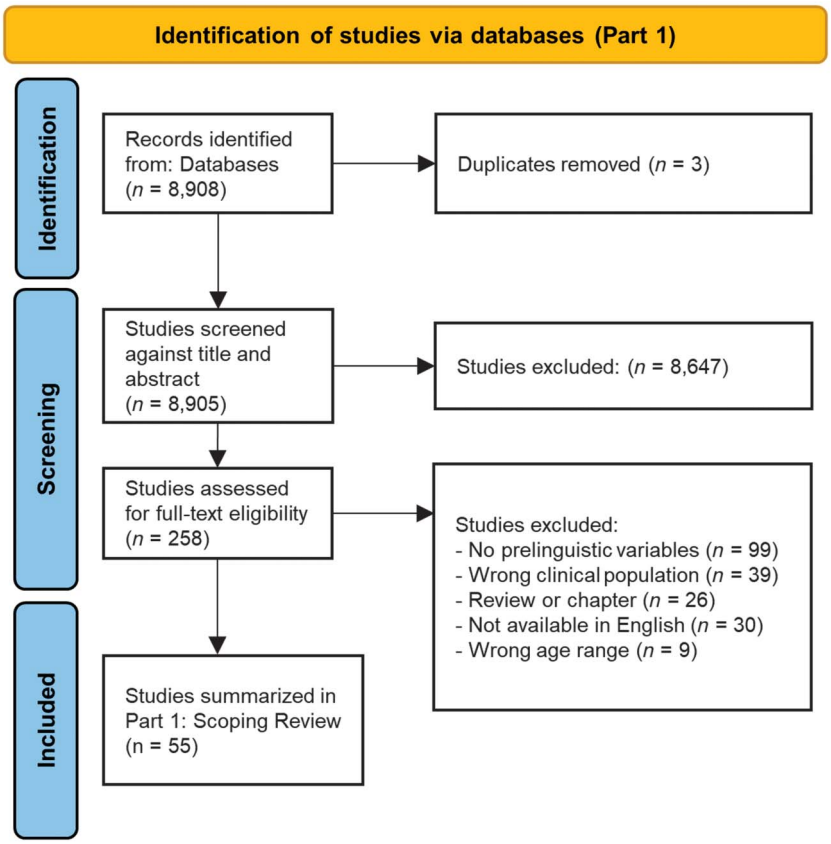
Research Method

The research methods used in each study are outlined in Table 3. Twenty-eight studies (51%) used a longitudinal research design. Forty-eight (87%) studies used observational methods. Across all studies reporting an observational session, the approximate mean length of session was 21 min (SD = 12.6).[2]

[1]All n counts indicate the number of papers.

[2]The approximate mean length of recording is estimated from available data across studies; individual articles varied in reporting means, ranges, or exact length of recordings in minutes.

Figure 1. Identification of studies via databases.



Forty-one studies (75%) used human judgment coding of infant vocalizations to tabulate perceptual laboratory calculations. Fifteen studies (27%) used caregiver-reported measures to estimate vocal milestone attainment using prospective or retrospective questionnaires. Caregiver questionnaires included the Infant Monitor of Vocal Production (Moore & Colyvas, 2018), the Checklist for the Development of Early Vocalizations (Lyytinen et al., 1996), and laboratory-developed checklists of infant behavior or questionnaires of developmental milestones. Six studies (11%) used acoustic instrumentation to quantify vocal parameters; three of these studies measured vocal duration, and three studies measured other acoustic variables such as acoustic dispersion, second formant (F2) frequency, abnormal phonation, fundamental frequency, and intonation contour. One study (2%) reported subjective judgments of acoustic variables typically measured instrumentally, for example, amplitude (e.g., loud/quiet) and pitch variations. One study used automated language analysis from the LENA software to calculate child vocalization counts.

Across the studies using observational measurement procedures, 18 studies (33%) measured the frequency of vocalizations produced in recordings. Sixteen (29%) measured the onset of specific prelinguistic vocal stages (e.g., canonical babbling). Fifteen studies (27%) calculated a ratio or proportion of vocalizations such as marginal babbling (slow consonant–vowel formant transitions) or canonical babbling (rapid, adultlike consonant–vowel transitions). Nine studies (16%) measured the rate of vocalizations (e.g., per minute, per 10-min interval).

With respect to the level at which vocalizations were studied, 33 studies (60%) quantified the well-formedness of syllable types, including 26 studies on canonical babbling (47%). Ten (18%) studies reported consonant diversity (i.e., number of different consonants). Six studies (11%) measured caregiver-reported information on vocal milestone emergence (e.g., estimated age of canonical babbling onset, presence/absence of vocalization types). Twenty-five studies (45%) measured vocalizations at the utterance level, and 15 studies (27%) measured vocalizations at the syllable level. Sixteen studies (29%) did not operationally define the level at which the terms "vocalizations" or "babbling" were segmented for measurement.

Table 2. Populations and methods of studies examining vocal production of infants at risk for speech motor involvement (Part 1).

Part 2	Paper	Clinical populations (*N*)	Age range	Research design	Research setting	Sampling method	Vocalization measures
**	Benassi et al. (2016)	Preterm (20)	12 months (corrected)	Cross-sectional analytic	Laboratory	Caregiver–infant interaction	Mean rate per 10 min of vocalization and babbling utterances
	Berger & Cunningham (1983)	Down syndrome (6)	6–24 weeks	Longitudinal prospective cohort	Home	Caregiver–infant interaction, still-face	Duration of vocal utterances
*	Bochner (1986)	Down syndrome (3), hydrocephalus (1), spina bifida (1)	2–17 months	Longitudinal case series	Hospital	Environment recording	Amount, pitch, and amplitude of vocalizations; number of different consonant types
*	Brown et al. (1986)	Preterm with IVH (21), preterm without IVH (12)	9–22 months (corrected)	Cross-sectional analytic	Laboratory	Caregiver–infant interaction, treatment, standardized assessment	Speech sound rating scale for speech age and speech quotient
*	Brown & Ruder (1995)	Preterm (20)	4 and 7 months	Longitudinal prospective cohort	Home	Caregiver–infant interaction	Number of vocalizations
	Cobo-Lewis et al. (1996)	Down syndrome (23)	0–120 weeks	Longitudinal prospective cohort	Laboratory	Caregiver–infant and lab staff interaction	Caregiver-reported onset of canonical babbling
	Dodd (1972)	Down syndrome (10)	9–13 months	Cross-sectional analytic	Laboratory	Infant alone	Amount and duration of vowel and consonantal utterances
*	Eilers et al. (1993)	Preterm (20)	0–60 weeks	Longitudinal prospective cohort	Home, laboratory, hospital	Caregiver–infant and lab staff interaction	Caregiver-reported onset of canonical babbling
	Fiani et al. (2021)	Down syndrome (3)	4 months	Single-subject ABAB	Home	Caregiver–infant interaction	Rate of speech and nonspeech sounds
*	Gec (2007)	"Pathology at birth" (hemorrhaging, hypoxia, or asphyxia) (30)	6, 9, 12, and 24 months	Longitudinal prospective cohort	Home, medical clinic	Milestone questionnaire, standardized assessment, lab staff interaction	Number of phonemes
*	Goggin et al. (1978)	Fetal malnutrition (23)	1, 2, 4, 6, 9, 12 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant interaction	Amount of babbling behavior
	Gunn et al. (1979)	Down syndrome (10)	4–19 months	Single-subject ABAB	Laboratory	Caregiver–infant interaction, still-face	Rate of vocalization
*	Hulme et al. (1989)	CP (8)	1.5–2.8 years	Single-subject AB	Home	Caregiver–infant interaction	Number of vowels, consonants, and nonspeech sounds
*	Jennische & Sedin (1999)	Preterm (284)	6.5 years	Retrospective case–control	Not stated	Milestone questionnaire	Caregiver report of absent baby babbling
*	Jensen et al. (1988)	"Perinatal risk group" (9)	6, 8, 11, 12, 14 months	Longitudinal prospective cohort	Not stated	Structured lab staff interaction	Number of different consonants in reduplicated and nonreduplicated syllables

(*table continues*)

Table 2. *(Continued).*

Part 2	Paper	Clinical populations (*N*)	Age range	Research design	Research setting	Sampling method	Vocalization measures
*	Largo et al. (1986)	Preterm (114), CP (21)	1, 3, 6, 9, 12, 18, 24 months	Longitudinal prospective cohort	Home and laboratory	Milestone questionnaire	Caregiver report of stages of vocal and language development
**	Levin (1999)	CP (8)	11–12 months	Case series	Home	Caregiver–infant interaction	Number of syllables and utterances, syllable type ratios, number of precursor vocalizations, percentage of syllables with a true consonant, number of vowel types, number of syllables per utterance
	Locatelli et al. (2021)	Down syndrome (105)	3–17 years	Retrospective survey	Medical clinic	Milestone questionnaire	Caregiver report of babbling milestone age of acquisition
**	Lohmander et al. (2017)	CP (4), chromosomal deletion syndrome (2)	9–21 months	Case series	Laboratory	Caregiver–infant interaction, milestone questionnaire	Number of different consonants; ratio of canonical utterances
	Lynch, Oller, Steffens, & Buder (1995)	Down syndrome (8)	2–12 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant and lab staff interaction	Duration of syllables, utterances, and prelinguistic phrases
	Lynch, Oller, Steffens, Levine, et al. (1995)	Down syndrome (13)	4–18 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant and lab staff interaction, milestone questionnaire	Age of canonical babbling onset; proportion of sessions with CBR $\geq$ 0.15
*	Marchman et al. (1991)	Brain injury (5)	11, 13, 21 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant and lab staff interaction	Number of vocalizations; number of syllables per utterance; percentage of consonant vocalizations by manner and place of articulation; proportion of vocalizations with true consonants
**	McCathren et al. (1999)	Down syndrome (4), preterm (4), failure to thrive (3) macrocephaly (1), microcephaly (1), neonatal meningitis (1)	17–34 months and 12 months postvisit	Longitudinal prospective case series	School	Structured lab staff interaction	Rate of vocalizations and vocalizations with consonants per minute
	McConkey & Martin (1984)	Down syndrome (10)	52–104 weeks	Longitudinal prospective case series	Home	Caregiver–infant interaction	Frequency count of vocalizations
*	Muñoz-Arbeláez et al. (2019)	Preterm (8)	0–12 months	Cross-sectional analytic	Not stated	Environment recording	Acoustic dispersion of babbling signals

(*table continues*)

Table 2. *(Continued).*

Part 2	Paper	Clinical populations (*N*)	Age range	Research design	Research setting	Sampling method	Vocalization measures
**	Nyman & Lohmander (2018)	Down syndrome (6), CP/suspected (6), chromosomal syndrome (2), brain malformation (1)	12–22 months	Cross-sectional analytic	Medical clinic	Caregiver–infant interaction	Percentage of children with a CBR > 0.15; percentage of children with consonant vocalizations by manner and place of articulation
**	Nyman, Strömbergsson, Lindström, et al. (2021)	Down syndrome (5), CP (4), chromosomal syndrome (2)	12–22 months, 5 years	Longitudinal case series	Laboratory	Caregiver–infant interaction	Number of different true consonants
*	Oller & Seibert (1988)	Down syndrome (8), seizures (10), "multiple disabilities" (8), microcephaly (4), motoric disorder (2), failure to thrive (3), hydrocephalus (3)	17–62 months	Cross-sectional analytic	Laboratory	Caregiver–infant–lab staff interaction	Ratio of canonical utterances
*	Oller et al. (1998)	"At risk for developmental disorders" (1,536)	10–22 months	Cross-sectional analytic	Laboratory, phone interview	Milestone questionnaire, caregiver–infant interaction	Rate of occurrence of late onset canonical babbling
*	Oller et al. (1999)	"High-risk population" (3,053)	10–12 months	Cross-sectional analytic	Laboratory, phone interview	Caregiver–infant interaction	Caregiver report of canonical babbling onset; proportion of infants with late onset canonical babbling
	Onnivello et al. (2021)	Down syndrome (74)	4–18 months	Incidence study without comparison	Laboratory	Standardized assessment	Caregiver report of vocal milestone attainment
*	Otapowicz et al. (2005)	CP (46)	3–16 years	Retrospective survey	Medical clinic	Milestone questionnaire	Caregiver report of cooing age of onset
	Pansy et al. (2019)	Prader-Willi (1)	27 weeks	Case study	Hospital	Lab staff interaction	Proportion of five levels of vocal complexity using SAEVD-R
	Peter et al. (2020)	Congenital galactosemia (5)	2–24 months	Randomized pilot trial	Laboratory	Intervention	Mean babbling levels; mean syllable structure levels
	Poulson (1988)	Down syndrome (3)	2.7–8.2 months	Single-subject ABAB	Laboratory	Caregiver–infant interaction, intervention	Rate of vocalizations per minute
*	Powell & Low (1983)	Failure to thrive (21)	3–32 months	Case series	Hospital	Environmental recording	Observed lack of or decreased vocalization
	Romano et al. (2020)	Down syndrome (19)	11–42 months	Longitudinal case series	Home	Structured caregiver–infant interaction, intervention	Rate of vocalizations per minute

(*table continues*)

Table 2. *(Continued).*

Part 2	Paper	Clinical populations (*N*)	Age range	Research design	Research setting	Sampling method	Vocalization measures
*	Ross (1985)	Preterm (46)	12 months	Cross-sectional analytic	Laboratory	Standardized assessment	Questionnaire item on Bayley Mental Scales: "Jabbers expressively"
	Rothbart & Hanson (1983)	Down syndrome (15)	6, 9, 12 months	Longitudinal prospective cohort	Home	Milestone questionnaire	Scaled score and range of Vocal Activity Scale of the Infant Behavior Questionnaire
**	Rvachew et al. (2005)	Preterm with BPD (13), preterm without BPD (9)	8, 12, 18 months	Longitudinal prospective cohort	Hospital	Caregiver–infant-lab staff interaction	Mean number of syllables per utterance; canonical syllable ratios; proportion of syllables by syllable structure type; number of consonants in repertoire; standard deviation of second formant frequencies; abnormal phonation ratio; consonant–vowel syllable duration
	Semenzin et al. (2021)	Angelman syndrome (10)	11–53 months	Cross-sectional analytic	Home	Environmental recording	Proportion of segments with canonical syllables
	B. L. Smith & Oller (1981)	Down syndrome (10)	0–9 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant interaction	Caregiver-reported age of onset of reduplicated babbling; frequency of occurrence of consonants and vowels
	L. Smith (1987)	Down syndrome (2)	12–23 months	Longitudinal case study	Home	Caregiver–infant interaction	Mean number of vocalizations per session
	B. L. Smith & Stoel-Gammon (1996)	Down syndrome (9)	6–25 months	Longitudinal case series	Laboratory	Caregiver–infant and lab staff interaction	Percentage occurrence of reduplicated and variegated utterances
	Sohner & Mitchell (1991)	Cri-du-Chat (1)	8–26 months	Case study	Laboratory	Caregiver–infant interaction	Rate of noncry vocalizations; proportion of intonation contours; mean fundamental frequency; onset of multisyllable babbling
	Steffens et al. (1992)	Down syndrome (13)	4–18 months	Longitudinal case series	Laboratory	Caregiver–infant and lab staff interaction	Mean quasivowel, full-vowel, marginal syllable, and canonical syllable ratios

(*table continues*)

Table 2. *(Continued).*

Part 2	Paper	Clinical populations (*N*)	Age range	Research design	Research setting	Sampling method	Vocalization measures
**	Stolt et al. (2012)	VLBW (32)	0–24 months	Longitudinal prospective cohort	Home, hospital	Milestone questionnaire	Mean age of acquisition for quasivowels, cooing, variation, and babbling
*	Suttora & Salerni (2011)	Preterm (16)	6–24 months (corrected)	Longitudinal case series	Laboratory	Caregiver–infant interaction	Mean babbling levels, frequency of vocal productions per minute
	Thiemann-Bourque et al. (2014)	Down syndrome (9)	9–11 months; 25–54 months	Case series	Home	Environmental recording	Child vocalization counts
**	Töröla et al. (2012)	Preterm (18)	0–9 months	Longitudinal prospective cohort	Hospital	Caregiver–infant–lab staff interaction	Milestone attainment based on 0.20 criterion across three stages of vocal development; number of missing skills
**	Ward et al. (2022)	CP (18)	6, 9, 12 months	Longitudinal prospective cohort	Home	Milestone questionnaire	Caregiver report of infant vocal complexity using Infant Monitor of Vocal Production
	Wolters et al. (2009)	Smith-Magenis syndrome (11)	5–34 months	Longitudinal case series	Laboratory	Standardized assessment	Descriptive behavior of occurrence of grunts, gurgles, squeals, and babbling
	Yoder et al. (2015)	Down syndrome (35)	18–27 months	Randomized clinical trial	Laboratory	Structured lab staff interaction, caregiver–infant interaction	Ratio of canonical syllable communication
	Zampini et al. (2022)	Sex chromosome trisomy (76)	18 months	Cross-sectional analytic	Laboratory	Caregiver–infant interaction	Mean number of vocalizations and babbling
**	Zuccarini et al. (2018)	Preterm (20)	6 and 12 months	Longitudinal prospective cohort	Laboratory	Caregiver–infant interaction	Mean rate of vocal production per 10 min

Note. IVH = intraventricular hemorrhage; ABAB = Baseline-Treatment-Baseline-Treatment; AB = Baseline-Treatment; CP = cerebral palsy; CBR = canonical babbling ratio; SAEVD-R = Stark Assessment of Early Vocal Development–Revised; BPD = bronchopulmonary dysplasia; VLBW = very low birth weight. *Quality appraised in Part 2. **Judged as high quality in Part 2.

Table 3. Research methods identified for Part 1.

Method	Variable	Total *N* = 55 (%)
Research setting	Laboratory	29 (53)
	Home	16 (29)
	Hospital/medical clinic	11 (20)
	Caregiver interview	2 (4)
	School/day care	1 (2)
	Not stated	3 (5)
Sampling method		
Unstructured observation	Parent–infant interaction	20 (36)
	Parent–infant and lab staff interaction	9 (16)
	Lab staff–infant interaction	5 (9)
	Environmental recording	5 (9)
	Infant alone play	1 (2)
Structured observation	Caregiver questionnaire	15 (27)
	Standardized assessment	5 (9)
	Structured lab staff interaction	3 (5)
	Structured PI interaction	1 (2)
	Still-face paradigm	2 (4)
Experimental	Single-subject design	4 (7)
	Randomized pilot trial	1 (2)
	Randomized controlled trial	1 (2)

Note. All percentages are calculated from the total number of studies (*N* = 55) included in Part 1. Reported counts do not sum to this total because many included more than one method or measure. Percentages do not sum to 100, and total counts do not sum to 55 because several studies involved more than one sampling method or setting. PI = parent-infant.

Discussion

Part 1 of our review synthesized the methodological landscape of a broad literature to understand the scope of research methods and measures used for the study of vocal characteristics in infants at risk for speech motor impairment. Specifically, we described the clinical populations and research methods used to study vocal characteristics of infants at risk for speech motor involvement.

We found a small percentage of studies (18%) explicitly examined infants based on a diagnosis or risk for CP. Almost half of the studies (45%) examined infants with Down syndrome, whereas the remaining studies examined other genetic conditions (16%), preterm infants (24%), and birth risk factors (20%) for CP. We expected to find a relatively small number of studies in CP because recruiting infants with CP in the first year of life is a complex task given that the diagnosis of CP may not occur until after 2 years of age. The average age range of studies examining infants with CP (11–26 months) was the oldest overall age range calculated compared to the other clinical groups; all other groups included at least one study that examined infants from birth. Recent advancements in the early detection of CP indicate that a diagnosis can now be made as young as 6 months of age (Maitre et al., 2023; Novak et al., 2017). Of note, the median year of publication for studies of infants with or at risk for CP was 2002. The substantial advancement in the potential for an earlier diagnosis of CP in the first year of life indicates a heightened prospect of future research in this area to recruit larger numbers of infants with a confirmed diagnosis of CP as young as 6 months.

The small number of studies explicitly examining vocal characteristics of infant with CP and the large number of studies examining mixed groups of heterogeneous clinical populations limit our ability to draw specific conclusions about methodological trends. Furthermore, the median publication year of studies of Down syndrome was 1995, indicating an even older body (compared to the CP study median year of publication: 2002) of work that can be only minimally applied when considering the sharp rise in technological advancements since the new millennium. Overall, we found large methodological differences across the 55 identified studies, a finding that parallels reviews of studies examining vocal characteristics in other populations (Lang et al., 2019; McDaniel & Gifford, 2020; L. Morgan & Wren, 2018). However, several commonalities emerged with respect to the sampling locations, methods, and measurement procedures that have potential to be applied to the future study of CP, expanded below.

Most studies (76%) identified in our review examined some form of mature consonant–vowel syllable production, although the measures and variables varied substantially, including the quantification of CBRs, consonant diversity, and parent-reported onset of canonical babbling. These findings highlight the longstanding attention to the development of canonical babbling as an indication of the command over the production of consonant–vowel syllable forms used in speech (Oller, 1978; Stark, 1980). Two studies calculated a mean babbling level or syllable structure level to summarize the overall production of syllable types into a single score (Paul & Jennings, 1992; Stoel-Gammon, 1989), and one study used the Stark Assessment of Early Vocal Development–Revised (Nathani et al., 2006), a coding protocol used to map vocalization types to their approximate stage of development. These alternative measures of canonical babbling may provide a more holistic view of developmental progress than the commonly used CBR, a measure that solely reflects the proportion of canonical syllables from all other noncanonical syllables and requires an arbitrary criterion to indicate attainment (C. C. Lee et al., 2018; Nyman, Strömbergsson, & Lohmander, 2021). Future studies should explore the utility of these alternative canonical babbling measures in longitudinal studies of emergent vocal and early speech development in infants with CP.

Forty-one (75%) studies identified in our search involved human coding of infant vocalizations. Human coding has long been considered the gold standard method to study vocal developmental characteristics (C. C. Lee et al., 2018). This method is theoretically justified given the perceptual salience of vocal categories; however, it is time- and cost-intensive (Nathani & Oller, 2001; Oller et al., 2021; Ramsdell et al., 2012). Also, over half of the studies (53%) were conducted in contrived laboratory settings where infant vocalizations are collected seminaturalistically during unstructured caregiver interactions (73%). These sampling methods are in line with the technological capabilities available to researchers across studies with a median publication year of 1999. However, substantial advancements in our ability to analyze infant vocalizations at much larger scales have emerged since this time.

Advancements in the development of smartphone technology and automated acoustic analysis software—such as the LENA recording software (Gilkerson & Richards, 2008; Gilkerson, Richards, Warren, et al., 2017)—can now be used to conduct dense sampling of infant vocalizations using day-long home audio recordings for naturalistic data collection. Only two studies (4%) used the LENA analysis software; one extracted the automatic child vocalization counts of infants with Down syndrome (Thiemann-Bourque et al., 2014), and another extracted 5-min segments from LENA recordings for hand coding of canonical and linguistic utterances in children with Angelman syndrome (Semenzin et al., 2021). Research using automated analysis software in language-based conditions has already indicated its potential for detecting biomarkers of later communication impairments in these populations (Oller et al., 2010; Pokorny et al., 2016, 2017, 2022; Warren et al., 2010). Reviews of that work reveal that automated detection methods can be just as accurate in detecting later impairment as hand-coding methods (Lang et al., 2019; Yankowitz et al., 2019). Additionally, only one study (2%) used smartphone recordings to analyze the acoustic dispersion of preterm infant babbling (Muñoz-Arbeláez et al., 2019). Smartphone applications for recording infant motor behaviors have also been shown to be feasible and acceptable to caregivers to track developmental progress (Kwong et al., 2019). Recently developed technology such as the Babbly application (Babbly.co, 2023) should be studied to support similar work in the vocal domain for these children.

Our scoping review highlights the paucity of research using automated acoustic analyses to detect speech motor involvement in any population, including CP. Only 13% of studies in our review used any acoustic analyses to measure infant vocalizations, and none of these examined infants with CP. Prior studies of acoustic parameters of typically developing infants and young children's speech also have yet to be applied to the study of vocal characteristics across populations. For example, studies on this topic have examined the development of "articulatory signatures" of mature consonant–vowel syllables (Singh & Singh, 2008) and the use of visual reinforcements to increase syllabic utterances using computer and smart technology applications (Daffern et al., 2020; Fell et al., 2003). This work has not been applied to the study of early vocal characteristics or intervention efforts aiming to enhance speech functioning. To this point, few studies used any experimental methods (just 11%), indicating a need to extend research efforts in building targeted interventions to support speech development in children with CP who use speech as their primary communication modality.

Although few studies utilized advanced analysis techniques, a somewhat larger percentage (27%) examined the onset of vocal milestones using caregiver-reported methods across populations and vocal stages, either prospectively (22%) or retrospectively (5%). Caregivers are known to be reliable reporters of their child's developmental milestone attainment, including canonical babbling (Lyytinen et al., 1996; Miller et al., 2017; Oller et al., 1998). Screening developmental milestones in clinical settings is a time- and cost-effective method for medical providers to quickly gauge the need for referral to speech-language specialists (Lipkin et al., 2020). Only one study in our review used a validated milestone questionnaire (Ward et al., 2022), notably in infants at risk for CP. All others

screened for vocal stage milestones using laboratory-developed measures or compared parent-reported ages of onset to typically developing age expectations. There are several other validated questionnaires that have not been applied to the study of vocal development in CP, such as the Ages & Stages Questionnaire (Squires & Bricker, 2009), the Vocal Development Landmarks Interview (Moeller et al., 2019), and the LENA Snapshot (Gilkerson, Richards, Greenwood, & Montgomery, 2017). Additional research is needed to compare these measures with hand-coded and/or automated detection software of acoustic parameters to examine their predictive validity for low-cost screening of speech motor involvement in CP and other populations.

Overall, our findings reveal an aging evidence base that has limited utility for making informed clinical decisions around the early diagnosis and prognosis of speech motor involvement in infants. Specifically, we found 55 studies across all conditions at risk for speech motor involvement with a median publication year of 1999, a median sample size of only nine participants, and major methodological differences that do not yet fully capitalize on the advanced technologies available to researchers today. The logistics behind recruiting children with relatively low-incidence conditions such as CP have historically limited researchers' ability to recruit large sample sizes. Recent advancements in the early diagnosis of CP hold promise for future recruitment of infants with a confirmed diagnosis of CP as young as 6 months of age. Ultimately, the study of infant vocalization is inherently complex, especially in the vastly heterogeneous population of CP, one that commonly presents with other comorbidities and concurrent speech and language impairments (a point further discussed in the Limitations of Parts 1 and 2 section below). It warrants the need for longitudinal designs, larger sample sizes, careful attention to the development of mature consonant–vowel syllable forms, and dense sampling using cutting-edge technology in order to detect vocal precursors of speech motor involvement. In doing so, we can further inform the diagnosis and prognosis of these children's speech development to support clinical decision making around the need to introduce augmentative and alternative communication (AAC) as early as possible.

Part 2: State of the Science on Infant Vocal Characteristics in CP

Method

Eligibility Criteria and Quality Appraisal

For Part 2 of our scoping review, we sought to synthesize the vocal characteristics reported across a subset of studies identified in Part 1 that examined infants with CP and with birth risk conditions for CP. Although quality review is uncommon in scoping reviews, we deemed it a necessary part of describing research findings that have the potential to be generalized to clinical contexts (Munn et al., 2018; Peters et al., 2020).

Prior to quality appraisal of studies identified in Part 1, we first excluded studies that only examined infants with genetic or chromosomal conditions (n = 25) because the vocal characteristics of these children may be unique to their genetic phenotype compared to infants who experienced birth complications likely to impact neurological development. After excluding these 25 studies, 30 studies examining infants with or at risk for CP underwent quality appraisal.

Quality appraisal was conducted on the remaining 30 studies using the Mixed Methods Appraisal Tool (MMAT; Hong et al., 2018) by the first and third authors independently. The MMAT was selected as a validated tool designed to appraise studies across five categories of research designs (quantitative randomized controlled trial, quantitative nonrandomized, quantitative descriptive, qualitative, and mixed methods). The 30 studies encompassed only two of these categories; we identified 22 quantitative nonrandomized (cohort, case–control, and cross-sectional analytic studies) and eight quantitative descriptive studies (surveys, case series, and case reports).

Using the MMAT, each reviewer assessed 100% of the 30 studies across the relevant methodological quality criteria for each design category. Reviewers answered each question as "yes" meets criteria, "no" does not meet criteria, or "unclear" where inadequate information was reported. Final independent quality ratings were based on the overall impression of quality from these responses to each question given that the MMAT discourages calculating an overall score from ratings of each criterion (Hong et al., 2018). Reliability was calculated using the Cohen's kappa indicating substantial agreement between reviewers, κ = .786 (Sim & Wright, 2005). A summary of the quality criteria collated through narrative synthesis is presented in Table 4.

A final quality judgment of low, moderate, or high quality was settled through consensus discussion. Only studies judged as high quality were selected to be synthesized as results. Individual study ratings across the two design categories are presented in Appendix B.

Results

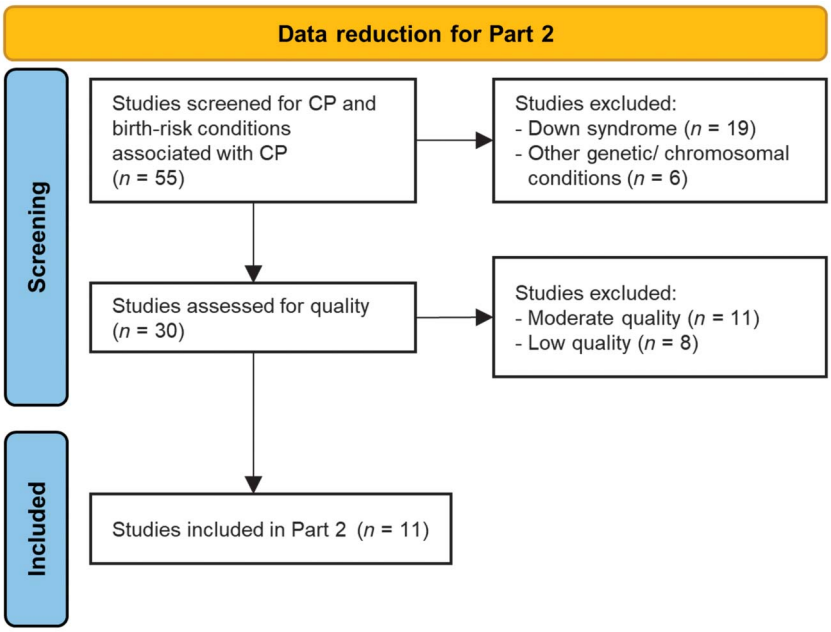
The data reduction flow diagram based on our eligibility criteria for Part 2 is shown in Figure 2. Of the 30 studies that underwent quality appraisal, only 11 (20%) studies met criteria as high quality for inclusion in Part 2.

Table 4. Quality appraisal criteria using the Mixed Methods Appraisal Tool.

Quantitative design type	Quality criteria	Number of studies that met criteria	Reasons studies did not meet criteria (i.e., received a "no" or "unclear" rating")
Screening questions (*N* = 30)	Are there clear research questions?	21	• Research questions unclear or not explicitly stated (*n* = 9)
	Do the collected data allow to address the research questions?	21	• Data collected did not reflect the variables or constructs as described in the research question or hypothesis (*n* = 9)
Nonrandomized studies (*n* = 22)	Are the participants representative of the target population?	16	• Target population was inadequately defined or undifferentiated across broadly described groups (*n* = 4) • Limited or no description of eligibility criteria (*n* = 2)
	Are measurements appropriate regarding both the outcome and intervention (or exposure)?	11	• Phonological or speech sound assessment as an inadequate measure of prelinguistic vocalization (*n* = 2) • "Vocalization" or "babbling" terms not operationally defined (*n* = 7) • Measure not well defined or justified for study of vocal production (*n* = 1) • Recording details of infant vocal production not adequately specified (*n* = 1)
	Are there complete outcome data?	19	• Reported data inadequately address the research questions (*n* = 1) • Missing data inadequately justified (*n* = 2)
	Are the confounders accounted for in the design and analysis?	14	• Within-group trait differences not adequately accounted for (*n* = 2) • Relevant demographic details not discussed or analyzed as potential confounds (*n* = 8) • Age- or sex-matching attempts not adequately discussed (*n* = 1)
	During the study period, is the intervention administered (or exposure occurred) as intended?	16	• Inadequate length of study period to measure change over time as hypothesized (*n* = 2) • Study used a single time-point, and change was not reported (*n* = 2) • Retrospective design using parent reported data affected validity of results (*n* = 1) • Intervention inadequately described as administered (*n* = 1)
Descriptive studies (*n* = 8)	Is the sampling strategy relevant to address the research question?	7	• Eligibility criteria was inadequate for target population of interest (*n* = 1)
	Is the sample representative of the target population?	7	• Target population was inadequately defined (*n* = 1)
	Are the measurements appropriate?	4	• "Vocalization" or "babbling" terms not operationally defined (*n* = 3)
	Is the risk of nonresponse bias low?	5	• Incomplete data for case series to answer research questions (*n* = 1) • Nonresponses not addressed or adequately discussed (*n* = 2)
	Is the statistical analysis appropriate to answer the research question?	6	• Retrospective design using parent reported data affected validity of results (*n* = 1) • Inadequate rationale for vocal behavioral findings as described (*n* = 1)

Note. Reviewers answered each question as "yes" meets criteria, "no" does not meet criteria, or "unclear" where inadequate information was reported. Final quality ratings were based on the overall impression of quality based on responses to each question and not a quantitative criterion of "yes" versus "no/unclear" ratings. The sum of reasons that did not meet criteria do not always sum to the total number of studies classified across each category because several studies did not meet criteria for more than one reason.

Figure 2. Data reduction for Part 2. CP = cerebral palsy.



The median year of publication for the 11 studies was 2016 (range: 1999–2022), and the median sample size was nine participants (range: 4–32). Detailed results on vocal characteristics and follow-up outcomes are presented by study in Table 5. A narrative synthesis is provided below across three categories of populations identified as CP, preterm and VLBW, and mixed clinical groups.

Cerebral Palsy

Only two studies examined a group of infants with CP or recruited for an explicit risk of CP (Levin 1999; Ward et al., 2022). Higher rates of marginal babbling and lower rates of canonical babbling were observed in both studies beyond expected ages of emergence.

Levin (1999) found that eight infants with CP between 11 and 12 months produced ~1 vocalization per minute (estimated from a range of total vocalizations reported across three half-hour sessions per infant), primarily low-back vowels, and only monosyllabic canonical utterances. Only 20% of their sample had reached the canonical babbling stage (as measured by a CBR criterion of 0.20), and all infants produced high rates of marginal syllables. Five of the eight participants produced any true consonants—more labials and velars than dental consonants—with an average of nine per session. More recently, Ward et al. (2022) examined the rate of vocal development in 18 infants at risk for CP and 18 typically developing infants using a parent-reported milestone questionnaire at 6, 9, and 12 months. The CP risk group showed a slower rate of vocal development compared to typically developing infants, with significantly lower performance at 9 and 12 months, particularly in the speech motor development domain, but not in a "social receptive" domain that was associated with phonation control and social interaction. Item-level analyses revealed marginal babbling contributed the greatest variance to the speech motor factor at 9 months and canonical babbling contributed the greatest variance at 12 months.

Preterm and Very Low Birthweight

Five studies examined vocal characteristics of preterm or VLBW infants across a range of ages between 0 and 24 months. The median sample size of these studies was 19 participants (M = 19, SD = 7.8). Across these studies, few significant vocal characteristic differences were observed among preterm and VLBW infants compared to full-term infants, further elaborated below across four methodological themes identified: vocal rate, canonical syllables, vocal stage emergence, and acoustic parameters.

Vocal rate. No significant differences were observed in the rate of vocalizations per minute between preterm and full-term infants. Preterm infants at or below 12 months of age demonstrated a mean rate of ~1.5 vocalizations per minute compared to full-term infants who produced ~2 per minute (Benassi et al., 2016; Töröla et al., 2012; Zuccarini et al., 2018). Decreasing rates of vocalizations were also observed in preterm infants and those with bronchopulmonary dysplasia (BPD), whereas full-term infants demonstrated higher or increasing rate of

Table 5. Results of studies examining vocal production of infants diagnosed with or at risk for cerebral palsy (Part 2).

Paper	Relevant clinical groups (*n*)	Age	Main findings	Follow-up outcomes	
				Age; measure	Outcome findings
Benassi et al. (2016)	Preterm (20)	12 months (corrected)	No differences between preterm and control groups in the frequency of vocalization, "babbling," or words at 12 months.	—	—
Levin (1999)	CP (8)	11–12 months	The onset of canonical babbling was delayed in six of eight participants. All had restricted phonetic repertoires and produced monosyllabic utterances only.	—	—
Lohmander et al. (2017)	"Neurodevelopmental disorder" group (10)	9–21 months	In a combined neurodevelopmental group, children produced a smaller number of different consonants and lower CBR than controls.	—	—
McCathren et al. (1999)	Preterm (4), failure to thrive (3) macro/microcephaly (2), neonatal meningitis (1)	17–23 months	In a combined clinical group, participants used 3.95 vocalizations per minute and 1.14 vocalizations with consonants per minute.	12 months after first session; number and rate of words	*M* = 13 different words; *M* = 0.66 different words per minute
Nyman & Lohmander (2018)	CP (4), Suspected CP (2), brain malformation (1)	12–22 months	In a combined neurodevelopmental group, a lower occurrence of babbling, consonant production, and CBRs were observed.	—	—
Nyman, Strömbergsson, Lindström, et al. (2021)	CP (4)	12–22 months	In a combined neurodevelopmental group, infants used three to eight different true consonants.	4:11–5:4 years; number of established consonants	The number of established consonants ranged from 5 to 11
Rvachew et al. (2005)	Preterm VLBW with BPD (13), preterm VLBW without BPD (9)	8, 12, 18 months	Infants with BPD produced smaller CBRs than controls. Infants without BPD caught up to controls by 18 months.	—	—
Stolt et al. (2012)	VLBW (32)	0–24 months	All participants were reported as producing quasivowels, cooing, variations in pitch/intensity, babbling, and first words at expected ages.	24 months; RDLS III, MCDI-Finnish	Age of babble/first words and rate of vocal development were all associated with later language performance
Töröla et al. (2012)	Preterm (18)	0–9 months	All vocal milestones (except cooing) were approximately 2 weeks delayed compared to controls.	—	—
Ward et al. (2022)	CP risk (18)	6, 9, 12 months	The rate of development of canonical babble was lower in the CP risk group compared to controls.	—	—
Zuccarini et al. (2018)	Preterm (20)	6 and 12 months	Manual exploration at 6 months predicted total vocal production at 12 months.	—	—

Note. Em dashes in "Follow-up outcomes" columns indicate these data were not reported or studied in the respective study. CP = cerebral palsy; CBR = canonical babbling ratio; VLBW = very low birth weight; BPD = bronchopulmonary dysplasia; RDLS-III = Reynell Developmental Language Scales–Third Edition; MCDI = MacArthur Communicative Development Inventories.

vocalization over time (Töröla et al., 2012; Rvachew et al., 2005). There were also no differences in the mean number of syllables per utterance (~1–2) between 8 and 18 months (Rvachew et al., 2005).

Canonical syllables. Preterm infants generally produced a smaller (often nonsignificant) proportion of canonical syllables, fewer canonical syllables (< 1) per minute, and lower number of consonants under 18 months

than full-term infants (Benassi et al., 2016; Rvachew et al., 2005). A smaller percentage of preterm infants produced any canonical syllables or reached the canonical babbling stage (as measured by a CBR > 0.2) at or before 12 months, indicating greater delays in reaching the canonical babbling milestone for preterm infants, especially those with BPD (Benassi et al., 2016; Rvachew et al., 2005). Although preterm infants with BPD had the lowest overall number of consonants, all preterm and full-term infants increased their number of consonants between 8 and 18 months. Notably, healthy preterm infants and full-term infants had a significantly larger vocabulary size at 18 months compared to preterm infants with BPD (Rvachew et al., 2005).

Vocal stage emergence. No differences were observed between preterm/VLBW and full-term infants in ages of onset for the phonation, cooing, expansion, reduplicated babbling, and first word stages of development (Stolt et al., 2012; Töröla et al. 2012). One study found preterm infants produced significantly fewer types of vocalizations during the cooing stage but not expansion stage, suggesting less variability in the early emergence of speech motor capabilities (Töröla et al., 2012). Of note, these two studies measured vocal stage emergence using different methods despite finding similar ages of onset across stages (parent-reported questionnaire vs. laboratory-based vocal coding), highlighting the potential for future work to validate parent-reported measures with laboratory human coding of infant vocal types to inform clinical screening of vocal precursors for speech motor involvement in these populations.

Acoustic parameters. Finally, one study examined acoustic parameter differences of preterm and full-term infant vocalizations. Rvachew et al. (2005) found no significant differences across acoustic measures of vowel space, abnormal phonation, and consonant–vowel syllable duration. However, the preterm infants with BPD consistently demonstrated the lowest measures overall. Specifically, from 8 to 18 months, they had the smallest vowel spaces (as measured by the standard deviation of the F2 frequency), smallest overall abnormal phonation ratios, and longest syllable durations with little change over time compared to healthy preterm and full-term infants.

Mixed Clinical Groups of Infants With Neurodevelopmental Conditions

Four studies examined mixed clinical groups of infants with neurodevelopmental conditions, including CP, CP risk factors, and genetic conditions, with a median of four different clinical groups included (M = 4.5, SD = 1.3). These studies reported a significantly smaller number of different consonants—particularly dental/alveolar plosives—produced by these clinical groups compared to typically developing controls (Lohmander et al., 2017; McCathren et al., 1999; Nyman & Lohmander, 2018). Lower rates of canonical babbling and a smaller percentage of infants reaching the canonical babbling stage were also noted (Lohmander et al., 2017; Nyman & Lohmander, 2018). One study observed a comparable number of vocalizations per minute (~4 per minute) as typically developing infants from recent studies on this topic (Oller et al., 2019, 2020).

Notably, one study (Nyman, Strömbergsson, Lindström, et al., 2021) was conducted as a follow-up study to Nyman and Lohmander (2018) and represents the only study we found that examined associations among early vocal production and later speech-language outcomes in children with neurodevelopmental disabilities (including CP). Specifically, Nyman, Strömbergsson, Lindström, et al. (2021) observed a moderate but nonsignificant correlation between infants' number of different consonants at 12–22 months and their percentage of consonants correct (PCC) at 5 years. Of the four children in their sample with CP, three (75%) were diagnosed with dysarthria and two (50%) has language impairments by 5 years, statistics that match prior studies reporting the prevalence of speech and language impairments in CP (Mei et al., 2020; Nordberg et al., 2013). Across their entire mixed clinical group, only 18% of their sample had age-appropriate speech or language abilities at 5 years, highlighting the clinical importance of identifying early and accurate predictors of these skills at the youngest possible ages.

Discussion

Part 2 of our scoping review sought to report the state of the high-quality evidence on vocal production in infants diagnosed or at risk for CP and speech motor involvement. Following a broad synthesis of the methods used across a broad range of populations at high risk for speech motor involvement (including infants with genetic or chromosomal conditions), we reduced our scope to studies examining infants with CP and those with known birth risk factors associated with CP. We identified 11 high-quality studies; two studies found delays in the onset of canonical babbling and protracted rates of marginal babbling in infants with CP, five studies found no or minimal vocal differences in the rate or onset of vocal stages for preterm/VLBW infants, and four studies found low vocal and canonical syllable rates in mixed clinical groups of infants. Large variability in the populations and methods of measurement were noted across these studies, a finding that parallels reviews of vocal development in other populations (Lang et al., 2019; McDaniel & Gifford, 2020; L. Morgan & Wren, 2018; Yankowitz et al., 2019). For this reason, our synthesis should be interpreted with caution. A discussion of these findings is provided below with respect to these trends to support the future direction of research on the early detection of speech motor involvement in CP.

Despite the methodological differences, we noted a preponderance of marginal syllables, low rates of canonical babbling and monosyllabic canonical utterances by 12 months, and small phonetic repertoires (less than six different consonants) through 24 months in infants with and at risk for CP across three studies (Levin, 1999; Lohmander et al., 2017; Nyman & Lohmander, 2018; Ward et al., 2022). Prior research on typical development has evidenced the consolidation of speech motor control via decreasing rates of marginal babbling and increasing rates of canonical babbling across the second half of the first year of life (Iverson & Thelen, 1999; Lewedag, 1995; Stark, 1980). Findings from this review suggest that the underlying neuropathology associated with a diagnosis of CP may also be observable in prelinguistic supraglottal articulation for at least some children. Specifically, we posit that high rates of marginal syllables during ages at which canonical syllables should predominate could potentially indicate early evidence of dysarthria (i.e., imprecise articulation). Future research should compare the associations among later levels of speech motor involvement with measures of vocal characteristics across longitudinal studies, including marginal and canonical babbling, consonant diversity, and specific canonical babbling characteristics (e.g., monosyllabic, reduplicated, and variegated babbling patterns).

Prior acoustic analyses of speech production in children with CP and speech motor involvement demonstrate smaller vowel spaces, longer vowel durations, reduced F2 slopes, and longer F2 transitions than those with no speech motor impairment (Allison & Hustad, 2018; J. Lee & Hustad, 2013; J. Lee et al., 2014). From our synthesis, Rvachew et al. (2005) also observed smaller vowel spaces and longer syllable durations in infants with multiple risk factors (i.e., preterm and BPD). These studies suggest that acoustic analyses of vowel space and consonant–vowel syllables (e.g., F2 slope and transition) may be used as an objective method of measurement to study the longitudinal development of supraglottal articulation (transitions from prelinguistic vocalization in infancy to speech into childhood) and offer insight into how these trajectories differ across children with and without speech motor involvement.

It is important to note that two of our identified studies observed noted differences in infants with multiple birth risk conditions compared to healthy preterm or full-term infants (Rvachew et al., 2005; Stolt et al., 2012). These findings may support prior research indicating that the risk of communication impairments in CP may be greater for infants born at-term and with higher birth weight, suggesting that prematurity alone is not a singular risk factor for communication impairments in CP (Pennington et al., 2020; Soares et al., 2017; Souza et al., 2019; Zhang et al., 2015). This is further corroborated by the studies that found minimal or no significant differences in the vocal characteristics between preterm and full-term infants (Benassi et al., 2016; Törölä et al., 2012; Zuccarini et al., 2018). However, these studies reported overall lower (~1–2) vocalizations per minute from both preterm and full-term groups of infants compared to recent studies (~4–5) using much larger samples of typically developing infants (Oller et al., 2019, 2020). Differences in the measurement of vocalizations may explain some discrepancies among studies; thus, we cannot definitively determine whether vocal rates differ among preterm and full-term infants based on extant literature. These findings underscore the notion that none of the vocal characteristics can yet be generalized to specific populations due to small sample sizes, a small number of studies identified, and considerable heterogeneity within the population of children overall. Regardless, the methodological and vocal findings synthesized in our review offer important insight into the future research directions needed to support the identification of vocal precursors to speech motor involvement in CP.

In regard to our discussion of multiple risk factors, future research should consider the extent to which specific birth risk factors and comorbidities may heighten the risk of greater levels of speech motor involvement in CP. For example, several studies have shown that infantile seizures and a higher number of comorbidities are associated with communication impairments in CP (Allison et al., 2023; Fluss & Lidzba, 2020; Hidecker et al., 2018). Thus, there is a need to examine differences in early vocal characteristics and subsequent speech production across conditions associated with CP that can be diagnosed in early infancy, such as seizures, periventricular leukomalacia, and hypoxic ischemic encephalopathy.

Only one study examined the association between infant vocal characteristics and later speech outcomes, finding a moderate (but nonsignificant) correlation between early consonant diversity and PCC at 5 years in a mixed clinical group of infants (Nyman, Strömbergsson, Lindström, et al., 2021). Two other studies found positive associations with later language performance of > 24 months, including with the age of babbling onset and the rate of canonical syllables at approximately 12 months (McCathren et al., 1999; Stolt et al., 2012). These studies reporting a mix of speech and language outcomes highlight an additional layer of complexity in this area of research. Prelinguistic vocal development is well established to reflect the interconnected development of both speech motor control and linguistic categories heard in their ambient environment (Laing & Bergelson, 2020; Lang et al., 2021; Oller, 2000). Moreover, many people with CP demonstrate speech motor and comorbid language impairments (Mei et al., 2015, 2020). Nyman, Strömbergsson, Lindström, et al. (2021) reported

both speech and language outcomes in their sample; however, their mixed clinical group of children limit our ability to draw any explicit conclusions about the predictive nature of early consonantal characteristics with later speech or language outcomes. Clearly, this finding necessitates the ongoing study of determining whether we have the potential to identify infant vocal precursors across a range of profiles of speech motor and language abilities in children with CP.

Limitations of Parts 1 and 2

The present review aimed to report the population and methodological landscape of research in infants at risk for speech motor involvement and the extent to which vocal characteristic findings revealed developmental differences specifically in those studies of infants with or at risk for CP. Throughout our search process, we noted some difficulty in identifying some populations as presenting with or without speech motor involvement given that many studies (especially older studies) often refer to communication broadly as "speech" when a closer look at their methods may reveal an interest in linguistic abilities. It is important to note that children with CP and other clinical populations discussed in Parts 1 and 2 of our review may present with concurrent speech motor disorders (dysarthria and childhood apraxia of speech [CAS]) and language impairment (Alvares & Downing, 1998; Chapman, 2017; Mei et al., 2015; Sanchez et al., 2019). In fact, several studies maintained a focus on language development despite their inclusion of participants also at risk for speech motor involvement, hence their inclusion in our review (Locatelli et al., 2021; McCathren et al., 1999; Romano et al., 2020). Our fair reliability observed in the title–abstract screening is a likely reflection of this problem; however, the full-text screening revealed substantial agreement beyond this point. Ultimately, the limited research reporting childhood outcomes limits our ability to connect vocal characteristic findings with either speech motor or language outcomes. These complexities indicate a substantial confound in our ability to generalize any findings to clinical practice, indicating a critical need for future research to work to tease apart potential precursors for the possible kinds of speech motor disorders and language impairment in CP.

Also, although idiopathic CAS was excluded from our search during Part 1, a recent review found only seven prior studies examining vocal development with respect to a later diagnosis of CAS and found notably lower vocal and canonical babbling rates compared to typically developing infants, indicating a need for more systematic study across all pediatric speech motor disorders (Overby & Highman, 2021). These and our present findings emphasize the gap in our understanding of the relationship between early vocal behaviors and speech outcomes across a variety of clinical populations. Also, we excluded 30 papers not available in English during our full-text screening in Part 1. Future studies could appraise and synthesize findings from these papers to expand our limited existing knowledge of vocal precursors of speech impairment in CP.

We eliminated children with Down syndrome and other genetic or chromosomal conditions in Part 2 because of their unique phenotype; however, a systematic review of findings across these conditions is clearly warranted to appraise the evidence base, particularly in Down syndrome. It is also worth noting that we encountered many studies during our screening phase in Part 1 that examined vocal development of infants with or at risk for other cognitive and intellectual disabilities and those with structural differences (i.e., cleft palate). Although these conditions were outside the scope of the present review, future studies could report the state of science on vocal production in these populations to support the early detection of vocal markers for their later outcomes as well.

Conclusions

Additional research examining vocal production of infants at risk for speech motor involvement, especially infants at risk for CP, is greatly needed. Methodological differences currently limit our ability to identify explicit vocal precursors of speech motor involvement and outcome associations across genetic and birth risk conditions. We noted broad delays in the emergence of canonical babbling beyond 12 months in infants prospectively identified for a diagnosis or risk of CP but mixed findings in studies examining infants with general birth risk factors (e.g., preterm and VLBW). Additional research is needed using larger sample sizes and modern technologies such as automated vocal detection software and advanced acoustic analyses to support the longitudinal study of relationships among vocal characteristics with later speech-language outcomes. This work is necessary to improve the early and accurate prediction of speech motor outcomes in infants at risk for speech motor involvement and to inform clinical decisions for implementing AAC in early intervention to support long-term communication outcomes in children with CP.

Author Contributions

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(Equal), Project administration (Lead), Resources (Equal), Software (Equal), Supervision (Lead), Validation (Lead), Visualization (Lead), Writing – original draft (Lead), Writing – review & editing (Lead). **Leslie Christensen:** Conceptualization (Supporting), Data curation (Equal), Formal analysis (Supporting), Investigation (Supporting), Methodology (Equal), Project administration (Supporting), Resources (Equal), Software (Equal), Validation (Supporting), Visualization (Supporting), Writing – original draft (Supporting), Writing – review & editing (Supporting). **Sydney Hayes:** Formal analysis (Supporting), Investigation (Supporting), Methodology (Supporting), Validation (Supporting), Visualization (Supporting), Writing – original draft (Supporting), Writing – review & editing (Supporting). **Katherine C. Hustad:** Conceptualization (Supporting), Formal analysis (Supporting), Funding acquisition (Supporting), Methodology (Supporting), Resources (Supporting), Supervision (Equal), Validation (Supporting), Visualization (Supporting), Writing – original draft (Equal), Writing – review & editing (Equal).

Data Availability Statement

This research protocol was preregistered on the Open Science Framework at https://osf.io/k8tnc. Data and supplemental material associated with this project are also available at https://osf.io/jf3u6/.

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Appendix A (p. 1 of 2)

Search Terms

Original Searches run on November 14, 2021

PubMed

("Cerebral Palsy"[mesh] OR "Down Syndrome"[mesh] OR "Chromosome Disorders"[mesh] OR "Apraxias"[mesh] OR "Speech Disorders"[mesh] OR mental-retardation[tiab] OR (apraxi*[tiab] OR dyspraxi*[tiab] OR dysarthri*[tiab] OR CP[tiab] OR trisomy-21[tiab] OR mongolism[tiab] OR oral-motor[tiab] OR infant-motor[tiab] OR CBR[tiab] OR MBL[tiab] OR SAEVD-R[tiab] OR VDLI[tiab] OR IMP[tiab] OR CAS[tiab] OR cerebral-pals*[tiab] OR ((motor[tiab] OR developmental*[tiab] OR neurodevelopmental*[tiab] OR speech[tiab] OR vocal[tiab] OR voice[tiab] OR chromosom*[tiab]) AND (disease*[tiab] OR disorder*[tiab] OR disabilit*[tiab] OR impairment*[tiab] OR delay[tiab] OR delays[tiab] OR deficit*[tiab] OR handicap*[tiab])) OR ((little*[tiab]) AND (disease*[tiab])) OR ((down[tiab] OR down-s[tiab] OR downs[tiab] OR prader-willi[tiab] OR cri-du-chat*[tiab] OR crying-cat[tiab] OR angelman*[tiab] OR 22q11*[tiab]) AND (syndrome*[tiab])))))

AND ("Language Development"[mesh] OR (((speech[tiab] OR language[tiab] OR vocal*[tiab] OR articulation*[tiab]) AND (development*[tiab])) OR babbl*[tiab] OR cooing[tiab] OR expansion-stage*[tiab] OR jargon*[tiab] OR prelinguist*[tiab] OR pre-linguist*[tiab] OR preverbal*[tiab] or pre-verbal*[tiab] OR prespeech*[tiab] OR pre-speech*[tiab]))

AND ("Infant"[mesh] OR infant*[tiab] OR infanc*[tiab] OR baby[tiab] OR babies[tiab] OR newborn*[tiab] OR new-born*[tiab] OR neonate*[tiab] OR neo-nate*[tiab] OR neonatal*[tiab] OR neo-natal*[tiab])

Scopus (Elsevier)

(TITLE-ABS-KEY(((mental W/3 retardation)) OR (cerebral W/3 pals*) OR ((oral OR infant*) W/5 (motor)) OR (trisomy-21 OR mongolism OR apraxi* OR dyspraxi* OR dysarthri* OR CP OR CBR OR MBL OR SAEVD-R OR VDLI OR IMP OR CAS OR ((motor OR developmental* OR neurodevelopmental* OR speech OR vocal OR voice OR chromosom*) W/5 (disease* OR disorder* OR disabilit* OR impairment* OR delay OR delays OR deficit* OR handicap*)) OR ((little*) W/3 (disease*)) OR ((down OR down-s OR downs OR prader-willi OR cri-du-chat* OR crying-cat OR angelman* OR 22q11*) W/3 (syndrome*)))))

AND (TITLE-ABS-KEY((((speech OR language OR **vocal OR articulation**) W/5 (development*)) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)))

AND (TITLE-ABS-KEY(infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*))

Web of Science (Science Citation Index Expanded [SCI-EXPANDED], Social Sciences Citation Index (SSCI), Conference Proceedings Citation Index–Science [CPCI-S], Conference Proceedings Citation Index–Social Science & Humanities [CPCI-SSH], Emerging Sources Citation Index [ESCI])

(TS = ((mental NEAR/3 retardation) OR ((cerebral) NEAR/3 (pals*)) OR ((oral OR infant*) NEAR/5 (motor)) OR (trisomy-21 OR mongolism OR apraxi* OR dyspraxi* OR dysarthri* OR CP OR CBR OR MBL OR SAEVD-R OR VDLI OR IMP OR CAS OR ((motor OR developmental* OR neurodevelopmental* OR speech OR vocal OR voice OR chromosom*) NEAR/5 (disease* OR disorder* OR disabilit* OR impairment* OR delay OR delays OR deficit* OR handicap*)) OR ((little*) NEAR/3 (disease*)) OR ((down OR down-s OR downs OR prader-willi OR cri-du-chat* OR crying-cat OR angelman* OR 22q11*) NEAR/3 (syndrome*)))))

AND (TS = ((((speech OR language OR vocal* OR articulation*) NEAR/5 (development*)) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)))

AND (TS = (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*))

CINAHL Plus with Full-Text (Elsevier)

((MH ("Cerebral Palsy" OR "Down Syndrome" OR "Chromosome Disorders+" OR "Apraxia+" OR "Speech Disorders+") OR TI ((mental N3 retardation) OR (cerebral N3 pals*) OR ((oral OR infant*) N3 (motor) OR trisomy-21 OR mongolism OR apraxi* OR dysprax* OR dysarthri* OR CP OR CBR OR MBL OR SAEVD-R OR VDLI OR IMP OR CAS OR ((motor OR developmental* OR neurodevelopmental* OR speech OR vocal OR voice OR chromosom*) N5 (disease* OR disorder* OR disabilit* OR impairment* OR delay OR delays OR deficit* OR handicap*))))) OR ((little*) N3 (disease*)) OR ((down OR down-s OR downs OR prader-willi OR cri-du-chat* OR crying-cat OR angelman* OR 22q11*) N3 (syndrome*))) OR AB ((mental N3 retardation) OR (cerebral N3 pals*) OR ((oral OR infant*) N3 (motor) OR trisomy-21 OR mongolism OR apraxi* OR dysprax* OR dysarthri* OR CP OR CBR OR MBL OR SAEVD-R OR VDLI OR IMP OR CAS OR ((motor OR developmental* OR neurodevelopmental* OR speech OR vocal OR voice OR chromosom*) N5 (disease* OR disorder* OR disabilit* OR impairment* OR delay OR delays OR deficit* OR handicap*))))) OR ((little*) N3 (disease*)) OR ((down OR down-s OR downs OR prader-willi OR cri-du-chat* OR crying-cat OR angelman* OR 22q11*) N3 (syndrome*))))) AND (MH ("Language Development") OR TI ((speech OR language OR vocal OR articulation) N5 (development*) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*) OR AB ((speech OR language OR vocal* OR articulation*) N5 (development*) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)) AND (MH "Infant+" OR TI (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate*) OR AB (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*)))

Revised and updated searches run on 5/12/22

Appendix A (p. 2 of 2)

Search Terms

PubMed

("Hypoxia Ischemia, Brain"[MeSH] OR "Stroke"[mesh] OR "Brain Injuries"[mesh] OR "Neuroinflammatory Diseases"[mesh] OR "Central Nervous System Vascular Malformations"[Mesh] OR "Infant, Extremely Premature"[Mesh] OR "Infant, Low Birth Weight"[Mesh] OR "Apgar Score"[mesh] OR (HIE[tiab] OR hypoxia*[tiab] OR hypoxic[tiab] OR stroke*[tiab] OR apoplex*[tiab] OR CVA[tiab] OR meningit*[tiab] OR encephal*[tiab] OR meningoencephal*[tiab] OR ventriculit*[tiab] OR arachnoidit*[tiab] OR neuroinflammatory[tiab] OR neuro-inflammatory[tiab] OR apgar[tiab] OR ((extreme*[tiab]) AND (prematur*[tiab] OR preterm*[tiab] OR pre-term*[tiab] OR preemie*[tiab])) OR ((low-birth[tiab]) AND (weight*[tiab])) OR ((cerebral[tiab] OR dural[tiab] OR cerebrovascular[tiab] OR cerebro-vascular[tiab] OR brain[tiab] OR central-nervous[tiab] OR CNS[tiab]) AND (accident*[tiab] OR damage*[tiab] OR injur*[tiab] OR infection*[tiab] OR inflammation*[tiab] OR malform*[tiab] OR anomaly[tiab] OR anomalies[tiab] OR abnormalit*[tiab] OR fistula*[tiab]))))) AND ("Language Development"[mesh] OR (((speech[tiab] OR language[tiab] OR vocal*[tiab] OR articulation*[tiab]) AND (development*[tiab])) OR babbl*[tiab] OR cooing[tiab] OR expansion-stage*[tiab] OR jargon*[tiab] OR prelinguist*[tiab] OR pre-linguist*[tiab] OR preverbal*[tiab] or pre-verbal*[tiab] OR prespeech*[tiab] OR pre-speech*[tiab]))

AND ("Infant"[mesh] OR infant*[tiab] OR infanc*[tiab] OR baby[tiab] OR babies[tiab] OR newborn*[tiab] OR new-born*[tiab] OR neonate*[tiab] OR neo-nate*[tiab] OR neonatal*[tiab] OR neo-natal*[tiab])

Scopus (Elsevier)

(TITLE-ABS-KEY(HIE OR hypoxia* OR hypoxic OR stroke* OR apoplex* OR CVA OR meningit* OR encephal* OR meningoencephal* OR ventriculit* OR arachnoidit* OR neuroinflammatory OR neuro-inflammatory OR apgar OR ((extreme*) AND (prematur* OR preterm* OR pre-term* OR preemie*)) OR ((low-birth) AND (weight*)) OR ((cerebral OR dural OR cerebrovascular OR cerebro-vascular OR brain OR central-nervous OR CNS) AND (accident* OR damage* OR injur* OR infection* OR inflammation* OR malform* OR anomaly OR anomalies OR abnormalit* OR fistula*))))) AND (TITLE-ABS-KEY(((((speech OR language OR vocal OR articulation) W/5 (development*)) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)))

AND (TITLE-ABS-KEY(infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*))

Web of Science (Science Citation Index Expanded [SCI-EXPANDED], Social Sciences Citation Index (SSCI), Conference Proceedings Citation Index–Science [CPCI-S], Conference Proceedings Citation Index–Social Science & Humanities [CPCI-SSH], Emerging Sources Citation Index [ESCI])

(TS = (HIE OR hypoxia* OR hypoxic OR stroke* OR apoplex* OR CVA OR meningit* OR encephal* OR meningoencephal* OR ventriculit* OR arachnoidit* OR neuroinflammatory OR neuro-inflammatory OR apgar OR ((extreme*) AND (prematur* OR preterm* OR pre-term* OR preemie*)) OR ((low-birth) AND (weight*)) OR ((cerebral OR dural OR cerebrovascular OR cerebro-vascular OR brain OR central-nervous OR CNS) AND (accident* OR damage* OR injur* OR infection* OR inflammation* OR malform* OR anomaly OR anomalies OR abnormalit* OR fistula*))))) AND (TS = ((((speech OR language OR vocal* OR articulation*) NEAR/5 (development*)) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)))

AND (TS = (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*))

CINAHL Plus with Full Text

((MH ("Hypoxia-Ischemia, Brain+" OR "Stroke+" OR "Brain Injuries+" OR "Central Nervous System Infections+" OR "Infant, Premature" OR "Infant, Low Birth Weight" OR "Apgar Score") OR TI (HIE OR hypoxia* OR hypoxic OR stroke* OR apoplex* OR CVA OR meningit* OR encephal* OR meningoencephal* OR ventriculit* OR arachnoidit* OR neuroinflammatory OR neuro-inflammatory OR apgar) OR AB (HIE OR hypoxia* OR hypoxic OR stroke* OR apoplex* OR CVA OR meningit* OR encephal* OR meningoencephal* OR ventriculit* OR arachnoidit* OR neuroinflammatory OR neuro-inflammatory OR apgar) OR TI ((extreme*) AND (prematur* OR preterm* OR pre-term* OR preemie*)) OR AB ((extreme*) AND (prematur* OR preterm* OR pre-term* OR preemie*)) OR TI ((low-birth) AND (weight*)) OR AB ((low-birth) AND (weight*)) OR TI ((cerebral OR dural OR cerebrovascular OR cerebro-vascular OR brain OR central-nervous OR CNS) AND (accident* OR damage* OR injur* OR infection* OR inflammation* OR malform* OR anomaly OR anomalies OR abnormalit* OR fistula*)) OR AB ((cerebral OR dural OR cerebrovascular OR cerebro-vascular OR brain OR central-nervous OR CNS) AND (accident* OR damage* OR injur* OR infection* OR inflammation* OR malform* OR anomaly OR anomalies OR abnormalit* OR fistula*))) AND(MH ("Language Development") OR TI ((speech OR language OR vocal OR articulation) N5 (development*) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*) OR AB ((speech OR language OR vocal* OR articulation*) N5 (development*) OR babbl* OR cooing OR expansion-stage* OR jargon* OR prelinguist* OR pre-linguist* OR preverbal* OR pre-verbal* OR prespeech* OR pre-speech*)) AND (MH "Infant+" OR TI (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate*) OR AB (infant* OR infanc* OR baby OR babies OR newborn* OR new-born* OR neonate* OR neo-nate* OR neonatal* OR neo-natal*)))

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MMAT Consensus Ratings for 7 Quantitative Descriptive Studies

Study	Screening questions		Nonrandomized studies					Quality judgment (high, mod., low)
	S1. Are there clear research questions?	S2. Do the collected data allow to address the research questions?	Are the participants representative of the target population?	Are measurements appropriate regarding both the outcome and intervention (or exposure)?	Are there complete outcome data?	Are the confounders accounted for in the design and analysis?	During the study period, is the intervention administered (or exposure occurred) as intended?	
Benassi et al. (2016)	Y	Y	Y	Y	Y	Y	Y	High
Nyman & Lohmander (2018)	Y	Y	Y	Y	Y	Y	Y	High
Rvachew et al. (2005)	Y	Y	Y	Y	Y	Y	Y	High
Stolt et al. (2012)	Y	Y	Y	Y	N	Y	Y	High
Töröla et al. (2012)	Y	Y	Y	Y	Y	Y	Y	High
Ward et al. (2022)	Y	Y	Y	Y	Y	Y	Y	High
Zuccarini et al. (2018)	Y	Y	Y	Y	Y	Y	Y	High
Brown et al. (1986)	N	Y	Y	U	Y	U	U	Mod.
Brown & Ruder (1995)	Y	Y	Y	N	Y	U	U	Mod.
Eilers et al. (1993)	Y	U	Y	U	Y	Y	Y	Mod.
Jensen et al. (1988)	Y	Y	Y	U	U	Y	Y	Mod.
Largo et al. (1986)	Y	Y	U	N	Y	Y	Y	Mod.
Marchman et al. (1991)	Y	Y	Y	N	Y	N	Y	Mod.
Oller & Seibert (1988)	U	Y	Y	Y	Y	Y	Y	Mod.
Oller et al. (1998)	U	Y	U	Y	N	Y	Y	Mod.
Oller et al. (1999)	N	Y	U	Y	Y	U	Y	Mod.
Ross (1985)	Y	Y	Y	N	Y	Y	Y	Mod.
Gec (2007)	N	U	U	N	Y	N	N	Low
Goggin et al. (1978)	Y	N	Y	N	Y	N	Y	Low
Hulme et al. (1989)	Y	N	N	N	Y	Y	U	Low
Jennische & Sedin (1999)	Y	U	Y	N	Y	N	N	Low
Muñoz-Arbeláez et al. (2019)	N	U	N	N	Y	N	U	Low

Note. Y = yes; N = no; U = unclear; Mod. = moderate.

MMAT Consensus Ratings for 22 Quantitative Nonrandomized Studies.

Appendix B (p. 2 of 2)

MMAT Consensus Ratings for 7 Quantitative Descriptive Studies

Study	Screening questions		Quantitative descriptive studies					Quality judgment (high, mod., low)
	S1. Are there clear research questions?	S2. Do the collected data allow to address the research questions?	Is the sampling strategy relevant to address the research question?	Is the sample representative of the target population?	Are the measurements appropriate?	Is the risk of nonresponse bias low?	Is the statistical analysis appropriate to answer the research question?	
Levin (1999)	Y	Y	Y	Y	Y	Y	Y	High
Lohmander et al. (2017)	Y	Y	Y	Y	Y	Y	Y	High
McCathren et al. (1999)	Y	Y	Y	Y	Y	Y	Y	High
Nyman, Strömbergsson, Lindström, et al. (2021)	Y	Y	Y	Y	Y	Y	Y	High
Suttora & Salerni (2011)	U	Y	Y	Y	U	Y	Y	Mod.
Otapowicz et al. (2005)	Y	N	Y	Y	N	N	N	Low
Powell & Low (1983)	N	U	N	Y	N	U	U	Low

Note. Y = yes; N = no; U = unclear; Mod. = moderate.