Examining Speech Production in Children with Cleft Palate with or without Cleft Lip: An Investigation of Characteristics related to Speech Articulation Skills

by

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ABSTRACT

Children with cleft palate with or without cleft lip (CP+/-L) often demonstrate disordered speech. Clinicians and researchers have a goal for children with CP+/-L to demonstrate typical speech when entering kindergarten; however, this benchmark is not routinely met. There is a large body of previous research examining speech articulation skills in this clinical population; however, there are continued questions regarding the severity of articulation deficits in children with CP+/-L, especially for the age range of children entering school. This dissertation aimed to provide additional information on speech accuracy and speech error usage in children with CP+/-L between the ages of four and seven years. Additionally, it explored individual and treatment characteristics that may influence articulation skills. Finally, it examined the relationship between speech accuracy during a sentence repetition task versus during a single-word naming task.

Children with CP+/-L presented with speech accuracy that differed according to manner of production. Speech accuracy for fricative phonemes was influenced by severity of hypernasality, although age and status of secondary surgery did not influence speech accuracy for fricatives. For place of articulation, children with CP+/-L demonstrated strongest accuracy of production for bilabial and velar phonemes, while alveolar and palatal phonemes were produced with lower accuracy. Children with clefting that involved the lip and alveolus demonstrated reduced speech accuracy for alveolar phonemes compared to children with clefts involving the hard and soft palate only.

Participants used a variety of speech error types, with developmental/phonological errors, anterior oral cleft speech characteristics, and compensatory errors occurring most frequently across the sample. Several factors impacted the type of speech errors used, including cleft type, severity of hypernasality, and age.

The results from this dissertation project support previous research findings and provide additional information regarding the severity of speech articulation deficits according to manner and place of consonant production and according to different speech error categories. This study adds information on individual and treatment characteristics that influenced speech accuracy and speech error usage.

DEDICATION

I dedicate my dissertation to my parents, Gary and Patricia Lien, and to my grandmother, Mary Lechtenberg. My parents have always modeled a can-do attitude and have acknowledged the process of learning as the priority in any educational endeavor. My grandma's frequent rosary prayers are probably the reason I am still intact after the intensity of completing a doctoral program. Thank you for being a constant source of love and support.

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CHAPTER 1

INTRODUCTION

Articulation disorders occur at a higher incidence in children with CP+/-L (Chapman et al., 2017), which may be attributed to early structural differences prior to cleft palate repair and/or persistent velopharyngeal insufficiency (VPI) after primary palate repair. Assessment of speech articulation skills during preschool and early school-age years is critical for monitoring skill development and for initiating speech intervention when indicated. The transition between preschool and early-school age (i.e., 4 to 7 years of age) is an especially important time to examine the development of articulation skills in children with CP+/-L due to developmental and structural factors that may influence achieving typical articulation skills.

Previous investigations of speech development in young children with cleft palate found that children with cleft demonstrate both cleft-related speech characteristics as well as more phonological speech errors than their peers without cleft (Klinto et al., 2014). At age five children with CP+/-L continued to present with reduced speech accuracy relative to their peers (Klinto et al., 2016). Numerous outcomes studies have reported on speech articulation skills in consecutive series of patients within a narrow age range (Britton et al., 2014; Sell et al., 2001; Willadsen et al., 2017; Wilson et al., 2019), with consistent reports of high usage of speech articulation errors within the samples. These studies have revealed that children with cleft palate continue demonstrating reduced speech articulation skills compared to age expectations; however, the targeted age ranges used in those previous studies have not allowed for analysis of how age predicts speech articulation skills in individuals with cleft during the preschool and early school-age years. The current study will attempt to address this gap in the literature by using a sample across the age range of 4;0 to 7;11.

Another factor that is known to impact speech articulation skills in children with CP+/-L is velopharyngeal insufficiency. Many research studies report the rate of symptoms associated with velopharyngeal insufficiency, including hypernasality and audible nasal emission when reporting speech articulation outcomes. However, these characteristics of velopharyngeal insufficiency have not been frequently examined as moderators of speech articulation outcomes (Lancaster et al., 2019), possibly due to lack of statistical power because of sample size limitations.

Speech accuracy according to manner of production has been examined in toddlers with cleft palate (Scherer et al., 2012); however, speech accuracy for different manner classes and according to place of consonant production has not been routinely reported in preschoolers and school-aged children with cleft. By examining speech accuracy according to specific sound classes, information on which sound classes continue to be more vulnerable to articulation errors within this later age range can be identified.

The purpose of this dissertation is to examine the speech articulation skills of children with CP+/-L between the ages of 4 and 7 years and to investigate factors that may influence speech skills in this clinical population. Understanding which factors predict improved speech articulation skills is important for identifying child characteristics that support better speech articulation skills. Specifically, chronological age and hypernasality will be investigated as predictors of speech accuracy and speech error usage. Additionally this study will investigate how speech accuracy differs according to manner of consonant production and place of consonant production in order to explore which sound classes are more vulnerable to articulation errors in preschool and school-aged children with CP+/-L.

CHAPTER 2

LITERATURE REVIEW

This literature review will summarize speech sampling material, the speech assessment protocol relevant to this study, and describe previous speech outcome studies.

Speech Material

The material used for speech sampling deserves important focus since speech material influences speech results (Klinto et al., 2011). According to the universal speech parameters outlined by Henningson and colleagues (2008), sentences should be constructed to meet the following criteria: (a) sentences to evaluate pressure consonants should be loaded with pressure consonants, (b) sentences should not include both pressure consonants and nasal consonants, and (c) sentences should include all vowels specific to the language used for assessment. Additionally, speech sampling should include at least one consonant from each manner class of high pressure consonants (i.e., stops, fricative affricates). Both voiced and voiceless sounds should be sampled for high-pressure consonants. Target consonants should be sampled in all positions of the word in which they appear. Henningson and colleagues (2008) suggested a minimum of 15 to 20 sentences in the speech sample for assessing hypernasality, nasal emission, and consonant production. Additionally, speech material recommendations indicate that non-target sounds should be glide or liquid phonemes, or should be as close as possible to the place of articulation as the target phoneme (Henningson et al., 2008).

The information outlined by the working group for universal speech parameters (Henningson et al., 2008) has provided an important foundation for this dissertation. The speech sample material used for the current project was the American English Sentence

Sample (AESS; Trost-Cardamone, 2012; See Appendix A). The AESS follows the universal parameters guidelines for how to sample speech at the sentence level. The AESS has also been used by the Americleft Speech Project (Chapman et al., 2018; Chapman et al., 2016).

Speech Assessment

Speech analysis used for the current project is based on work conducted using the Cleft Audit Protocol for Speech – Augmented (CAPS-A) and the subsequent Cleft Audit Protocol for Speech – Augmented – Americleft Modification (CAPS-A-AM; Chapman et al., 2016). The purpose of developing the CAPS-A (John et al., 2006) was to establish a tool for recording and reporting speech outcomes that demonstrated validity and reliability. The tool provides indication of individuals requiring furthering intervention for speech articulation and/or resonance concerns. While the CAPS-A tool was developed for the purpose of completing a national audit of speech outcomes for individuals with cleft palate, it was deemed appropriate for use in the clinical setting as well (John et al., 2006).

The CAPS-A yielded ratings on the following speech parameters: intelligibility/distinctiveness, voice, hypernasality, hyponasality, audible nasal emission, nasal turbulence, and grimace. The CAPS-A was later modified for use in the Americleft Speech Project (i.e., Cleft Audit Protocol for Speech – Augmented - Americleft Modification; CAPS-A-AM; Chapman et al., 2016). The equal-appearing interval scales for hypernasality, hyponasality, and audible nasal emission that were used for this dissertation were based on the scales used for the CAPS-A and also adopted by the CAPS-A-AM. Hypernasality is rated on a five-point equal appearing interval scale, while both hyponasality and audible nasal emission are rated on a three-point equal appearing interval scale. Further description of the scalar points for the speech parameters of hypernasality, hyponasality, and audible nasal emission is provided in the method section.

In addition to the speech parameters described above, the CAPS-A recorded consonant production. The CAPS-A provided useful categories of speech errors, later termed cleft speech characteristics (CSC), which are common in individuals with history of cleft palate or velopharyngeal insufficiency. These main categories included anterior oral CSC, posterior oral CSC, non-oral CSC, and passive CSC (John et al., 2006). The CAPS-A-Americleft Modification also used these four error categories for CSCs, although some of the discrete error types within the categories had been updated since the development of the CAPS-A. Based on the CAPS-A-AM, the anterior oral CSC included the error categories of dentalization, lateralization, and palatalization. While these anterior oral errors are the most frequently-occurring anterior CSCs, these specific error types do not capture other anterior errors seen in individuals with cleft palate, including bilabial place of articulation for fricatives or reversed labiodental (i.e., dentolabial) phonemes that may occur in the presence of dental hazards to precise articulation. Therefore for the current study, an additional error group termed "other anterior oral CSCs" was included within the anterior oral CSCs to capture these anterior oral errors that occurred.

Posterior oral CSCs as described by CAPS-A-AM included double articulation of oral phonemes (i.e., production of /t/ and /k/ simultaneously) or backing to velar or uvula place of production. Non-oral CSCs included pharyngeal articulation, glottal articulation, nasal fricatives, or double articulation including glottal stops (CAPS-A-AM). The three categories of anterior oral CSCs, posterior oral CSCs, and non-oral, or compensatory, CSCs, are all considered active speech articulation errors. These placement errors are the result of misarticulations occurring regardless of velopharyngeal function that would not change with surgical intervention (Harding & Grunwell, 1998). Active speech characteristics are different from passive speech characteristics, which are considered obligatory consonant errors secondary to velopharyngeal insufficiency. The category of passive CSCs, as described by CAPS-A-AM, includes weak pressure consonants, nasalized voiced consonants, or nasal realization of pressure consonants. Passive CSCs are structural speech errors that successful velopharyngeal management would eliminate, while active CSCs require speech therapy for remediation (Harding & Grunwell, 1998).

Previous work on the CAPS-A and the CAPS-A-AM have provided significant contributions towards the speech error coding of cleft speech characteristics for the current project. However, the consonant production analysis used by these tools does not sufficiently address developmental and/or phonological speech errors that children with cleft palate may use in addition to CSCs. The CAPS-A-AM includes a binary response to indicate if other speech errors (e.g., fronting, stopping, gliding, cluster reduction) are present in the child's speech production. Prevalence of developmental and phonological speech error use is not adequately captured by the CAPS-A-AM speech analysis.

In order to describe developmental speech errors (i.e., speech errors that are appropriate for the child's age) and phonological speech patterns (i.e., speech error patterns used to simplify speech production), the Khan-Lewis Phonological Analysis, Third Edition (KLPA-3, Khan & Lewis; 2015) Core Processes list was referenced. The KLPA-3 includes 12 core processes that are observed frequently in the speech of young children and are considered developmental in nature. The core processes includes speech errors related to manner of consonant production, place of consonant production, reduction, and voicing differences. Specifically, manner processes include deaffrication, gliding of liquids, stopping of fricatives and affricates, stridency deletion, and vocalization. Place processes include palatal fronting and velar fronting. Reduction processes include cluster simplification, deletion of final consonants, and syllable reduction. Voicing processes include final devoicing and initial voicing. These core processes from the KLPA-3 supported the development of speech error codes for developmental/phonological speech errors for this study.

Previous speech outcomes projects

Eurocleft. One of the earliest projects aimed at comparing outcomes using an intercenter collaboration model was the Eurocleft Cohort Study (Grunwell et al., 2000). The Eurocleft Speech project evaluated the consonant production of children between the ages of 11 and 14. Most of the children in this cohort were judged to have acceptable speech, which would be expected for children in their early adolescence. Since the Eurocleft speech project, significant methodological advances have occurred in the collection and evaluation of speech outcomes. One of the difficulties of the Eurocleft Speech Project was evaluating speech outcomes for speakers of five different languages. Despite the cross-linguistic differences, the older age of the patient cohort, and the lack of accepted protocol for assessing speech outcomes, the Eurocleft team helped to initiate and provide motivation to continue evaluating cleft palate speech in future projects.

Cleft Standards Advisory Group Audit. Speech outcomes from the Cleft Standards Advisory Group (CSAG) Audit in the United Kingdom provided information regarding history of speech therapy, nasality, consonant errors, and speech intelligibility for a sample of five-year-old children and a sample of twelve-year-old children (Sell et al., 2001). Speech intelligibility remained a large concern for these cohorts, with 19% of 12-year-olds and 51% of 5-year-olds presenting with speech that was “different enough to provoke comment, unintelligible to strangers, or impossible to understand” (Sell et al., 2001, p. 34). Despite the amount of time spent in speech therapy as reported by the study, school-aged children continued demonstrating reduced speech intelligibility than what would be expected in non-cleft peers.

Benchmarking speech standards in the United Kingdom. A project to develop national standards for processes and outcomes for the care of cleft palate speech was conducted by Britton and colleagues (2014) in response to the poor outcomes reported by the CSAG project. This project included a phase of development of the process and outcome standards as well as a second phase to pilot national data against these standards. Initially, speech outcomes for five-year-olds were selected as a focus due to the importance that normal speech has on educational and social aspects for these children as they enter school.

The second phase of the project examined how national data for five-year-old children (*n*=1110) with all cleft types (UCLP, BCLP, CPO) compared to the outcome standards over a three year period (2001-2003). The first speech outcome standard stated that over 50% of five-year-old children will demonstrate normal speech, which reflects the care provided by the multidisciplinary cleft team. Results for the first benchmark indicated 48% of children demonstrated normal speech across the three years studied, which was not significantly different from the outcome standard. The first outcome appeared to be set at an appropriate level based on these results. The second speech outcome was related to the quality of the surgeon's care, stating that 70% of children should demonstrate no evidence of a structural speech problem and should not have had a secondary surgery or fistula repair for speech. Unfortunately the percentage of children demonstrating no evidence of structural concerns or history of secondary repair was significantly lower than the outcome standard over the course of three years. For the years 2002 and 2003, about 36% of children had speech problems that were structural in nature. The third and final outcome standard is a measure of the care provided by the SLP, audiologist, and surgeon. It states that 50% of children should have no cleft-related articulation problems that require therapeutic or surgical intervention. Results indicated the outcome across three years was significantly higher than the benchmark, suggesting the benchmark point should be increased.

The arduous work conducted by the specialist SLPs who evaluated the speech samples contributed to the largest audit of cleft palate speech that had been previously completed. The audit provided information on what cleft teams in the United Kingdom should expect for speech outcomes for five-year-old children. Additionally, the authors highlighted important methodological issues pertaining to perceptual evaluation of speech outcomes and conducting a clinical audit.

Cleft Care UK. The Cleft Care UK study was conducted to study the effects of centralizing cleft care based on poor outcomes reported in the Cleft Standards Advisory Group Audit. Between the CSAG project and the CCUK project, the number of cleft centers was reduced from 57 centers to only 11 centers. This reduction in centers increased the volume of new patients seen by each operating cleft group annually

(Persson et al., 2015). The Cleft Care UK project was a cross-sectional study of five-year-old children with unilateral cleft palate. To measure speech outcomes, the CAPS-A tool was used. In the CCUK project, the five-year-old children presented with lower prevalence of hypernasality and with a lower prevalence of non-oral and passive speech patterns relative to the CSAG project (Sell et al., 2015). An additional improvement was observed for 'normal' speech intelligibility, with 56.3% of the children in the CCUK project presenting with intelligible speech as compared to 19.6% of the children in the CSAG audit. Interestingly, in both the CCUK and CSAG, nearly 20% of children demonstrated significant concerns regarding reduced speech intelligibility.

Americleft Speech Project. The Americleft Speech Project compared speech outcomes for 5-year-old and 6-year-old children across three different centers. The speech assessment protocol was developed based on modifications of the CAPS-A protocol. These modifications included replacing the sentence stimuli used by the UK with the American English Sentence Sample (AESS; Trost-Cardamone, 2012). Additionally, some of the parameters and their definitions were modified for the Americleft project (Chapman et al., 2016).

Chapman and colleagues (2017) presented results of a benchmarking project that compared the speech of kindergarteners with cleft palate to their non-cleft peers. There were significant differences between the groups of children for speech acceptability, hypernasality, and number of errors on the Goldman-Fristoe Test of Articulaton-2. Additionally, 31% of children with cleft palate demonstrated suspected velopharyngeal dysfunction and 68% of children with cleft palate used atypical articulation errors (Chapman et al., 2017). Children with cleft palate used a greater number of speech errors on both the single-word naming task and the sentence repetition task (Wilson et al., 2019). Children with cleft palate used more errors across all four error categories of the CAPS-A-AM (i.e., anterior oral CSCs, posterior oral CSCs, non-oral CSCs, and passive CSCs; Wilson et al., 2019).

Scandcleft. The Scandcleft Project reports speech articulation outcomes for 5-year-olds with unilateral cleft lip and palate who participated in the randomized controlled trials that assessed surgical protocols. The project included a series of three trials comparing a common surgical procedure (i.e., lip and soft palate closure at three to four months; hard palate closure with vomer flap at 12 months) to three local procedures, including the following: (a) delayed hard palate closure at 36 months of age; (b) single-stage palate closure protocol with lip closure at 3-4 months and closure of hard and soft palate at 12 months; (c) lip closure at 3-4 months combined with a single-layer closure of the hard palate using vomer flap, followed by soft palate closure at 12 months. These local surgical procedures are compared to the common procedure in Scandcleft Trials 1, 2, and 3 respectively. When using accurate consonant production as an outcome, children who received earlier repair of the hard palate in the first trial of the Scandcleft project achieved higher percentage of consonants correct (when /s/-errors were disregarded) when compared to the children who received later closure of the hard palate (Willadsen et al., 2017). However, there were no other significant differences for consonant proficiency based on surgical method (Willadsen et al., 2017).

Summary. The projects discussed above represent the diligent efforts of numerous individuals to learning more about cleft-related speech outcomes and to improving the care provided to patients with cleft lip and palate. The initial projects reviewed provided motivation and support to other working groups' contributions to collecting outcomes. This dissertation study follows the methodology of the Americleft Speech Project in its use of speech sampling material (i.e., AESS) and its use of the CAPS-A-AM speech assessment protocol. This project aims to extend the speech analysis completed by the Americleft Speech Project to also include error analysis of developmental/phonological errors and to examine speech accuracy according to manner and place of articulation.

CHAPTER 3

METHOD

Data Source

Clinical data from speech-language evaluations conducted by the Barrow Cleft & Craniofacial Center (BCCC) have been entered in a Research Electronic Data Capture (REDCap) database for quality improvement and quality assurance (Harris et al., 2009). Speech-language evaluation data have been entered by a speech-language pathologist from the cleft palate team (K.L.) or by research assistants trained in extracting data from the speech-language evaluation protocol. This *BCCC CL/CP Registry for QI/QA* is the data source for this study.

Additionally, during each speech-language evaluation conducted at BCCC using a standardized protocol based on the Americleft Speech clinical protocol (Cordero et al., 2018), an audio recording of the individual's speech sample is obtained as standard of care. The audio-recorded speech samples are also used as data for this study.

Participants

This study examined the speech production of children with cleft palate with or without cleft lip (CP+/-L) between the ages of 4;0 and 7;11. Initially this project aimed to evaluate the speech production of children aged 3;0 through 7;11; however, review of audio samples revealed that many 3-year-olds were unable to complete the speech sample protocol for the American English Sentence Sample (AESS; Trost-Cardmone, 2012). Only four three-year-old children were able to complete production of the AESS. Therefore, the age range was adjusted to 4;0 through 7;11 in order to represent the age range of children able to complete the AESS sentence repetition task.

Inclusion and Exclusion Criteria

For individuals from the *BCCC CL/CP Registry for QI/QA* to be included in this study, the following criteria were met: (a) individual has diagnosis of cleft palate with or without cleft lip, (b) individual is between the ages of 4;0 and 7;11 at the time of the speech evaluation encounter, (c) individual has received a speech evaluation conducted at the Barrow Cleft & Craniofacial Center (BCCC) between February 2016 and June 2019, which included an audio-recorded speech sample, (d) the audio-recorded speech sample material was the American English Sentence Sample (See Appendix A), (e) the individual completed production of the AESS in English.

Individuals from the *BCCC CL/CP Registry for QI/QA* were excluded for the following reasons: (a) diagnosis of submucous cleft palate, (b) diagnosis of a genetic syndrome or another congenital anomaly in addition to cleft palate, (c) individual was younger than 4;0 or was older than 7;11, (d) during the AESS sentence repetition task, the participant did not produce at least 53 of the 59 total target phonemes, which represented approximately 90% of the target phonemes, (e) the individual's audio sample contained significant environmental noises that impeded hearing the child's sound production.

The data pull from the *BCCC CL/CP Registry for QI/QA* yielded 114 participants who had received at least one speech-language evaluation at BCCC between February 2016 and June 2019. Available cases were reviewed against inclusion and exclusion criteria, and ineligible cases were removed from the data set. A summary of excluded cases is provided in Table 1.

Table 1. Reasons for exclusion from study.

Reason for exclusion	Number

	Excluded
Participant did not have diagnosis of non-syndromic CP+/-L	*n*=26
Participant was outside of the age range of 4;0 to 7;11	*n*=6
Participant did not have an audio file available in BCCC records	*n*=3
Audio file contained significant environmental noises that impeded assessment of speech production from audio sample	*n*=4
Speech sample was the 3-year-old version of the sentences in place of the American English Sentence Sample	*n*=19

For individuals who received more than one speech-language evaluation between February 2016 and June 2019, the earliest speech-language evaluation that included production of the AESS was used for analysis.

Description of Sample

Fifty-six children with non-syndromic cleft palate with or without cleft lip (CP+/-L) were included in this study. There were 31 females and 25 males. Mean age was 5.47 years, with a range from 4.01 to 7.97 years and a standard deviation of 1.17 years. Twenty-five participants had clefts of the palate only, while 31 participants had clefts of the lip and palate (23 participants were diagnosed with unilateral cleft lip and palate; eight participants were diagnosed with bilateral cleft lip and palate). Nine individuals had a diagnosis of Pierre Robin sequence. Fifteen children had history of secondary palate surgery.

Materials

Sentence Repetition Task

During the clinical encounter, a speech sample was conducted that included a sentence repetition task using the American English Sentence Sample (AESS; Trost-Cardamone, 2012; Sentence sample is available in Appendix A). The AESS consists of 24 sentences that are each loaded with a specific target sound. Importantly, the sentences separate pressure consonants (i.e., stop consonants, fricative consonants, and affricate consonants) from nasal consonants to prevent any confounding co-articulatory effects when assessing speech. Additionally, the only phonemes included in sentences other than the target phoneme of interest are low-pressure phonemes or are similar in place of production (Trost-Cardamone, 2012). The AESS follows the guidelines for universal parameters of speech as outlined by Henningsson and colleagues (2008). The child's repetition of the AESS was audio-recorded as a part of standard of care. The audio-recorded sentence repetition task was the data source for analyzing speech articulation skills during a sentence repetition task.

Single-word Articulation

Single word articulation testing was conducted during the clinical encounter at BCCC using the Goldman Fristoe Test of Articulation, Third Edition (GFTA-3) Sounds-in-Words subtest. The GFTA-3 is a valid and reliable measure of single-word articulation skills using a standardized, norm-referenced assessment (Goldman & Fristoe, 2015). Consonants produced in error were transcribed using narrow phonetic transcription by one of three speech-language pathologists at BCCC in order to calculate the raw score. The speech-language pathologist converted the raw score to a standard score and percentile rank using the GFTA-3 administration manual. The GFTA-3 raw score, standard score, and percentile rank were reported in the clinical evaluation documentation, which were entered in the *BCCC CL/CP Registry for QI/QA*. Since the standardized articulation assessment using the GFTA-3 was conducted during the clinical encounter, there was not a video or audio recording available for performing reliability analysis on speech articulation for single word productions. The speech articulation skills assessed in single-word productions should be interpreted as clinical data representing speech functioning as assessed by speech-language pathologists with expertise in assessing cleft speech characteristics.

Perceptual Assessment of Speech

A perceptual evaluation of speech parameters including hypernasality, audible nasal emission, hyponasality, and voice was conducted using the Cleft Audit Protocol for Speech-Augmented-Americleft Modification (CAPS-A-AM; Chapman et al., 2016; John et al., 2006). The CAPS-A-AM uses an equal-appearing interval scale for rating the parameters of hypernasality, audible nasal emission, hyponasality, and voice. Information on these scales is available in Table 2. Perceptual ratings of speech parameters were conducted using the audio-recorded speech sample that included the sentence repetition task, serial counting from 1-20 and from 60-70, and a brief spontaneous conversation sample.

Table 2. Description of speech parameters from the CAPS-A-AM protocol.

Speech Parameter	Brief description of speech parameter	Information on rating scale	Rating scale anchors
Hypernasality	An abnormal increase in nasal resonance	Five-point scale	0 – None 1 – Minimal hypernasality, slight increase in nasal resonance 2 – Mild hypernasality, which is evident on high vowels 3 – Moderate hypernasality, which is evident on all vowels 4 – Severe hypernasality which is evident on all vowels and voiced consonants
Audible Nasal	An abnormal or	Three-point	0 – None

Emission	inappropriate air escape through the nose during the production of high pressure consonants	scale	1 – Occasionally Present 2 – Frequently Present
Hyponasality	reduction or absence of nasal resonance on nasal phonemes	Three-point scale	0 – None 1 – Mild denasalization of nasal consonants 2 – Marked denasalization of nasal consonants and adjacent vowels
Voice	voicing classified as normal, or of clinical concern	Three-point scale	0 – Normal 1 – Mild Concern 2 – Moderate/Severe Concern

Design

This is a retrospective cross-sectional study examining the speech articulation skills of individuals with CP+/-L. The goal of this cross-sectional study is to examine the speech accuracy and speech error usage in children with CP+/-L between the ages of four and seven.

Procedure

Barrow Cleft & Craniofacial Center Speech Evaluation Procedure

The speech samples conducted during speech-language evaluations at BCCC were audio-recorded as standard of care in following the Parameters for Evaluation and Treatment of Individuals with Cleft Lip/Palate or Other Craniofacial Differences (American Cleft Palate-Craniofacial Association, 2018). Audio-recordings were obtained using Audacity software (Mazzoni, 1999) and with a Blue Microphone –Snowflake, Blue Microphone – Snowball, or Blue Microphone – Snowball Ice (Blue, 2018). Audio recordings were saved to a secured network drive on the hospital network using the individual's medical record number and date of speech encounter as the file name per hospital policy for audio file storage.

Transcribing and Analyzing Speech Articulation Skills for Sentence Repetition Task

Using the audio-recorded samples of the sentence repetition task, the full sentences were transcribed by the first author (K.L.) using narrow phonetic transcription using the Extensions to the International Phonetic Alphabet (extIPA) symbols (International Phonetic Alphabet, 2015). The transcription of target sounds for the initial word position, medial word position, and final word position within the AESS were coded and entered in an Excel workbook for further analysis. The codes for speech errors are available in Table 3. The cleft speech characteristic codes are based on work reported by John et al. (2006), and the phonological error analyses are based on the Core Processes analyzed by the Khan-Lewis Phonological Analysis, Third Edition (Khan & Lewis, 2015). For each of the main speech error category codes, speech error subcategory codes were assigned to analyze speech articulation errors with greater precision. All coding of speech errors was performed by the first author (K.L.). When coding speech errors, the following error categories were added to describe errors observed in the sample that could not be classified according to the cleft speech characteristic codes or the Khan-Lewis Phonological Analysis codes: (a) ingressive speech errors, (b) phonological nasal substitutions, (c) other developmental errors such as (th) errors. Voicing differences were transcribed using diacritics but were not coded as speech errors.

Table 3. Speech error coding system.

Code	Speech error category	Subcategory code	Examples of speech errors
1	Correct production of phoneme	N/A	N/A

2	Anterior oral CSC	21	Dentalization
2	Anterior oral CSC	22	Lateralization
2	Anterior oral CSC	23	Palatalization, Mid-dorsum palatals
2	Anterior oral CSC	24[a]	Bilabial fricatives, dentolabial placement
3	Posterior CSC	31	Double articulation (alveolar with velar production)
3	Posterior CSC	32	Backed to velar or uvular articulatory placement
4	Non-oral compensatory error	41	Pharyngeal articulation
4	Non-oral compensatory error	42	Glottal articulation
4	Non-oral compensatory error	43	Active nasal fricatives
4	Non-oral compensatory error	44	Double articulation with glottal stop
5	Passive CSC	51	Weak or nasalized consonants
5	Passive CSC	52	Nasal realization of plosives
5	Passive CSC	53	Gliding of fricatives or affricates
6	Developmental/phonological errors related to manner	611	Deaffrication
6	Developmental/phonological errors related to manner	612	Gliding of liquids
6	Developmental/phonological errors related to manner	613	Stopping of fricatives and affricates
6	Developmental/phonological errors related to manner	614	Stridency Deletion
6	Developmental/phonological errors related to manner	615	Vocalization
6	Developmental and phonological errors related to place	621	Palatal fronting
6	Developmental and phonological errors related to place	622	Velar fronting
6	Developmental and phonological errors related to place	623[b]	"Other" place of articulation errors, including (th) placement errors
6	Developmental and phonological errors related to reduction	631	Cluster simplification
6	Developmental and phonological errors related to reduction	632	Deletion of final consonant
6	Omission of Initial or Medial Consonants – Phonological error related to reduction	633	Complete consonant omission of initial or medial consonant
6	Developmental and phonological errors related to reduction	634	Syllable reduction
7	Ingressive Articulation Errors	N/A	Inhilatory fricatives; Non-pulmonic clicks
8	Phonological Nasal Substitution	N/A	Nasal substitution for stop, fricative, affricate, or liquid phoneme

a. A category was added for “other anterior oral cleft speech characteristics” to include bilabial fricative substitutions and dentolabial articulatory placement. This category is not an original error type listed in the CAPS-A-AM protocol.
b. A category was added for “other developmental/phonological errors related to place of articulation” since placement errors for (th) could not be accounted for by KLPA-3 core phonological processes.

An Excel workbook was created to calculate percentage of consonants correct (PCC) for total consonants as well as according to manner of consonant production and place of consonant production. PCC for manner of articulation was calculated for the following phoneme classes: nasals, glides, stops, fricatives, affricates, and liquids. PCC for place of articulation was calculated specifically for bilabial phonemes, alveolar phonemes, palatal phonemes, and velar phonemes. PCC for place of articulation was not computed for labiodental or interdental phonemes since those articulatory places were not of interest for the current study. The workbook used the COUNTIF function applied to specific cells to count the number of phonemes produced accurately for each manner and place of articulation. The number of phonemes produced accurately for each sound and manner class was divided by the number of phonemes the child attempted for each sound and manner class. If the child did not attempt production of a target phoneme (i.e., missing data), it was assigned a code of 999 and was excluded from the consonants attempted during the sentence repetition task. All PCC results described in this study are based on the phonemes attempted by the child, so as to not penalize the child for missing data.

Missing data.

Phonemes were coded as missing and were excluded from speech analysis for the following reason: (a) the child made a grammatical error resulting in use of wrong target word (e.g., “her” for target “she), (b) the child omitted the target word and was not prompted to repeat the target, (c) the clinician modeled only a portion of the sentence instead of the full sentence, (d) environmental noise or the clinician’s speaking covered up the child’s production of the target phoneme. Any speech sample with more than 6 missing phonemes was excluded from the project. Thirty-one participants (55.4% of the sample) produced all 59 target phonemes of the AESS. Seventeen participants (30.4% of the sample) only had one or two missing phonemes. Eight participants (14.3% of the sample) had between three and six missing phonemes.

Analyzing Speech Articulation Skills for Single-Word Production

The GFTA-3 raw score was obtained from the *BCCC CL/CP Registry for QI/QA*. Using the GFTA-3 raw score, the percentage of consonants correct (PCC) was calculated for the single word articulation test. Since there are 141 consonants produced on the GFTA-3 and the raw score represents the number of consonant production errors, the raw score was first subtracted from the total number of consonants on the assessment in order to determine the number of consonants produced correctly. Then the consonants produced correctly were divided by total number of consonants in order to yield percentage of consonants correct (PCC; Shriberg et al., 1997; Shriberg & Kwiatkowski, 1982). Single-word naming tasks represent the child’s best attempt at articulation and are routinely used in clinical assessments, which is why PCC was calculated for the GFTA-3 Sounds-in-Words subtest.

Analyzing Perceptual Speech Parameters

Perceptual ratings of hypernasality, audible nasal emission, hyponasality, and voice were performed by the first author (K.L.) with blinding to patient identity. This rater has been trained in perceptual speech assessment using the CAPS-A-AM scale by one of the speech raters from the Americleft Speech project (K.N.C.) through listener training courses conducted at BCCC. Additionally, the rater has conducted co-evaluations with the other BCCC speech-language pathologists (K.N.C., J.W.) for additional training and for calibration. Only individual medical record number and date of clinical speech-language evaluation encounter were available to the speech-language pathologist performing the blinded perceptual ratings.

Listening Procedures

Phonetic transcription and perceptual ratings of speech parameters were performed using over-the-ear headphones. The samples were played using the automatic settings in Windows Media Player. Recorded speech samples were replayed as many times as was necessary in order to make perceptual ratings for the speech parameters of interest or to transcribe the target phonemes.

Phonetic Transcription Reliability

Inter-rater reliability of phonetic transcription was performed for a random selection of 20% of the samples (*n*=12) by a student trained in IPA transcription specific to cleft speech characteristics. Point-by-point agreement was assessed for manner and place of production, and discrepancies in transcriptions were resolved through consensus listening. If consensus listening could not resolve disagreements in transcription, another speech-language pathologist trained in assessing cleft palate speech (J.W.) transcribed target phonemes and served as the “tiebreaker” for resolving disagreements. To calculate the percentage of agreement, the total number of agreements was divided by the total number of agreements plus disagreements, and this quotient was multiplied by 100.

Interobserver agreement was 76% between the first author and the trained undergraduate student.

Intra-rater reliability for phonetic transcription was performed for a random selection of 20% of the samples (*n*=12). Approximately six to eight weeks separated initial phonetic transcription and the secondary transcriptions for reliability Point-by-point agreement for place and manner of production was assessed. Percent agreement for intrarater reliability was 92%.

Reliability for Perceptual Parameters of Speech

Inter-observer reliability was completed for 20% of the speech samples (*n*=12) by two speech-language pathologists trained in the CAPS-A-AM protocol (J.W., K.N.C). The speech samples included in the reliability analysis were selected randomly. Prior to performing reliability, the Barrow Cleft & Craniofacial Center speech-language pathology team completed baseline ratings. Then listener calibration was conducted for 10% of the speech samples (*n*=6). Inter-observer reliability was completed for three speech parameters included in the CAPS-A-AM protocol, including hypernasality, audible nasal emission, and voice. Weighted kappa was not computed for the speech parameter of hyponasality, since none of the speech samples included participants with hyponasality. Weighted kappa statistics were calculated for hypernasality, nasal emission, and voice. The assigned weights followed the methodology described by Chapman and colleagues (2016; 1.00 for exact agreements; 0.89 for ratings that differed by 1 severity level; 0.56 for ratings that differed by 2 severity levels; 0 for ratings that differed by more than 2 severity levels). The weighted kappa statistic for each parameter and a description of the strength of agreement is presented in Table 4.

Table 4. Inter-rater reliability for CAPS-A-AM speech parameters.

Speech Parameter	Weighted Kappa	Strength of Agreement
Hypernasality	0.83	Very good agreement
Audible Nasal Emission	0.75	Good agreement
Voice	0.60	Moderate agreement

Intrarater reliability of perceptual ratings of hypernasality, nasal emission, and voice was performed for 20% of the samples (*n*=12). Weighted kappa was not computed for the speech parameter of hyponasality, since none of the speech samples included demonstrated hyponasality. Approximately two to three weeks separated initial ratings of perceptual parameters of speech and the secondary ratings for reliability. Weighted kappa statistics were calculated according to the weights reported by Chapman and colleagues, (2016), and these are reported in Table 5.

Table 5. Intra-rater reliability for perceptual speech parameters.

Speech Parameter	Weighted Kappa	Strength of Agreement
Hypernasality	0.81	Very good agreement
Audible Nasal Emission	0.75	Good agreement
Voice	0.64	Good agreement

Proposed Analyses

Aim one.

The first broad research aim was to examine speech accuracy in children with CP+/-L according to total speech accuracy as well as for manner and place of articulation. To address the first research aim, descriptive statistics were performed to provide means and standard deviations by consonant manner classes and by consonant place classes. Total speech accuracy was presented and classified according to PCC speech severity ratings. It was hypothesized that children with CP+/-L would demonstrate highest speech accuracy for the manner classes of glide, nasal, and stop phonemes, while their lower speech accuracy would be for production of fricatives, affricates, and liquids. In terms of place of production, it was expected that speech accuracy would be significantly higher for bilabial production and velar production relative to alveolar and palatal place of production.

Additionally, correlational analyses were performed to assess the linear relationships among manner classes and among different places of articulation. Visual analysis of scatterplots was conducted to determine which manner classes demonstrated linear relationships and which place classes had linear relationships prior to conducting analyses. It was expected that each class of high pressure consonants (e.g., stops, fricatives, and affricates) would be significantly correlated. For place of articulation, it was expected that alveolar and palatal place of articulation would be significantly correlated. Additionally, factors that may impact speech accuracy were explored.

Analysis of variance was used to examine how speech accuracy for fricative consonants differed according to severity of hypernasality. It was hypothesized that children with more severe hypernasality would have lower speech accuracy for fricatives.

Analysis of variance was also used to examine how speech accuracy for alveolar consonants differed according to cleft diagnosis. It was expected that individuals with clefting involving the lip, alveolus, and palate would demonstrate lower speech accuracy than children with clefts of the palate only.

Finally, a linear regression analysis was used to determine whether severity of hypernasality, history of secondary surgery, and age were significant predictors of total percentage of consonants correct during connected speech production. This regression analysis was repeated for speech accuracy for fricative consonants, since this class of phonemes requires high intraoral pressure for production, which may be impacted by velopharyngeal insufficiency. It was expected that hypernasality would significantly predict total speech accuracy and speech accuracy for fricatives, with children demonstrating hypernasality having reduced speech accuracy relative to children without hypernasal speech. It was also hypothesized that older age would predict higher speech accuracy. Individuals who have received secondary surgery would be expected to demonstrate higher speech accuracy.

Aim two.

The second broad research aim examined speech error usage in children with CP+/-L. Descriptive analyses were used to determine the frequency of speech errors produced across speech error categories (i.e., anterior oral articulation errors, posterior articulation errors, non-oral compensatory articulation errors, passive speech characteristics and developmental/phonological errors). It was hypothesized that younger children would demonstrate more posterior articulation errors, non-oral compensatory articulation errors, and phonological/developmental errors, while older children would demonstrate more anterior oral articulation errors.

To explore the influence of individual and treatment characteristics on the frequency of speech error usage, analysis of variance was used to examine differences in frequency of speech errors for categorical variables (i.e., cleft type, severity of hypernasality, history of secondary surgery), while regression analysis was used to examine differences in speech error usage for continuous variables (i.e., age). Given the number of statistical tests being conducted, a Bonferroni correction was be applied to reduce the rate of Type I error. The Bonferroni correction set the alpha level at 0.005.

Aim three. The third broad research aim examined speech accuracy for a single-word naming task compared to a sentence repetition task. Descriptive statistics were performed to examine the mean speech accuracy of children with CP+/-L on a sentence repetition task and for single-word production and to explore the variability in speech accuracy for these contexts. A correlational analysis was performed in order to examine the relationship of speech accuracy across contexts of production (i.e., single-words versus connected speech). It was expected that children with CP+/-L will have higher mean speech accuracy for single word productions than in connected speech. Additionally, proportion of the sample presenting with speech articulation disorders according to standardized testing was examined.

CHAPTER 4

RESULTS

Speech Accuracy: Aim One Results

Total Speech Accuracy

In this sample of children with CP+/-L between the ages of 4 and 7 years, the mean speech accuracy for total consonants was 69.25, with a range of 27.59 to 100 (S.D. = 17.70). According to Shriberg and Kwiatkowski's PCC severity of involvement metric, the mean speech accuracy for this sample represents a mild-to-moderate speech disorder (1982). Table 6 presents frequency of speech disorder severity levels according to Shriberg and Kwiatkowski's metric. The largest percentage of participants was classified as having a mild-to-moderate speech disorder, which represents total speech accuracy ranging from 65% and 84.99%.

Table 6. Frequency of speech disorder severity based on total speech accuracy.

Severity of Speech Disorder Classification (Range of PCC)	Number of Participants (Percentage of Sample)
Mild (85 – 100%)	10 (17.9%)
Mild-Moderate (65 – 84.99%)	24 (42.9%)
Moderate-Severe (50 – 64.99%)	12 (21.4%)
Severe (Less than 50%)	10 (17.9%)

Speech Accuracy by Manner of Articulation

Children with CP+/-L demonstrated differences in speech accuracy, measured by percentage of consonants correct, across manners of articulation. Descriptive statistics for different manners of speech articulation are reported in table 7. Speech accuracy was highest for nasal and glide consonants, and speech accuracy was lowest for affricate and liquid consonants. There was a large decrease in speech accuracy between stop consonants and fricative consonants. Additionally, the variability of speech accuracy was largest for fricative, affricate, and liquid consonants.

Table 7. Descriptive statistics for speech accuracy by manner of consonant production.

Manner of Production	Mean (S.D.)
Nasal Consonants	92.32 (11.48)
Glide Consonants	97.32 (11.36)
Stop Consonants	86.85 (16.11)
Fricative Consonants	57.39 (26.62)
Affricate Consonants	46.34 (39.03)
Liquid Consonants	43.75 (36.07)

Descriptive statistics by age group for speech accuracy by manner of consonant production is presented in Table 8. Variability of speech accuracy for different manner classes was large across the age range of participants.

Table 8. Descriptive statistics for manner of speech production by age group.

Manner of Production	4-year-old (*n*=22) Mean (S.D.)	5-year-old (*n*=16) Mean (S.D.)	6-year-old (*n*=10) Mean (S.D.)	7-year-old (*n*=8) Mean (S.D.)
Nasals	94.16 (8.80)	91.29 (12.73)	91.25 (10.29)	90.63 (17.36)
Glides	93.18 (17.56)	100 (0.00)	100 (0.00)	100 (0.00)
Stops	86.83 (15.60)	89.79 (9.03)	84.63 (20.02)	83.83 (24.10)
Fricatives	51.69 (26.82)	56.61 (21.76)	63.07 (28.79)	67.57 (32.60)
Affricates	36.36 (36.32)	63.44 (32.70)	44.00 (44.02)	42.50 (47.13)
Liquids	35.23 (29.54)	40.63 (39.66)	57.50 (37.36)	56.25 (41.73)

The relationships among speech accuracy for different manner classes were examined. It was hypothesized that nasal phonemes and glide phonemes would not have linear relationships with high-pressure consonants (e.g., stop consonants, fricative consonants, and affricate consonants) since children with CP+/-L acquire nasal and glide phonemes earlier in development since velopharyngeal insufficiency does not limit acquisition of these manner classes. Additionally, it was hypothesized that nasal and glide phonemes would not demonstrate linear relationships with liquid phonemes given the different ages of acquisition according to normative data. Visual inspection of scatterplots with linear best fit lines as well as with smoothed loess lines were used to establish whether linear relationships were present. Correlation analysis was only conducted for the relationships that were linear, including the following pairs of manner classes: (a) stop phonemes with nasal phonemes, (b) stop phonemes with fricative phonemes, (c) fricative phonemes with affricate phonemes, (d) fricative phonemes with liquid phonemes, and (e) affricate phonemes with liquid phonemes. Outliers were not excluded from analyses. The Pearson product moment correlations are presented in table 9 with a summary of each relationship. Nine relationships between speech manner classes were not examined, as they did not demonstrate monotonic relationships. Glide consonants did not demonstrate monotonic relationships with any other manner class (i.e., nasals, stops, fricatives, affricates, liquids). Additionally, nasal consonants were not systematically related to accuracy for fricatives, affricates, or liquids. Finally, stop consonants did not have a linear relationship with liquid phonemes.

Table 9. Summary of relationships between different manners of articulation with linear relationships.

Pairings between different manners of articulation	Pearson Product Moment Correlation Statistic	Description of Relationship
Nasals with Stops	r=0.24	Nonsignificant relationship
Stops with Fricatives	r=0.56*	Strong positive correlation
Fricatives with Affricates	r=0.59*	Strong positive correlation
Fricatives with Liquids	r=0.47*	Moderate positive correlation
Affricates with Liquids	r=0.45*	Moderate positive correlation

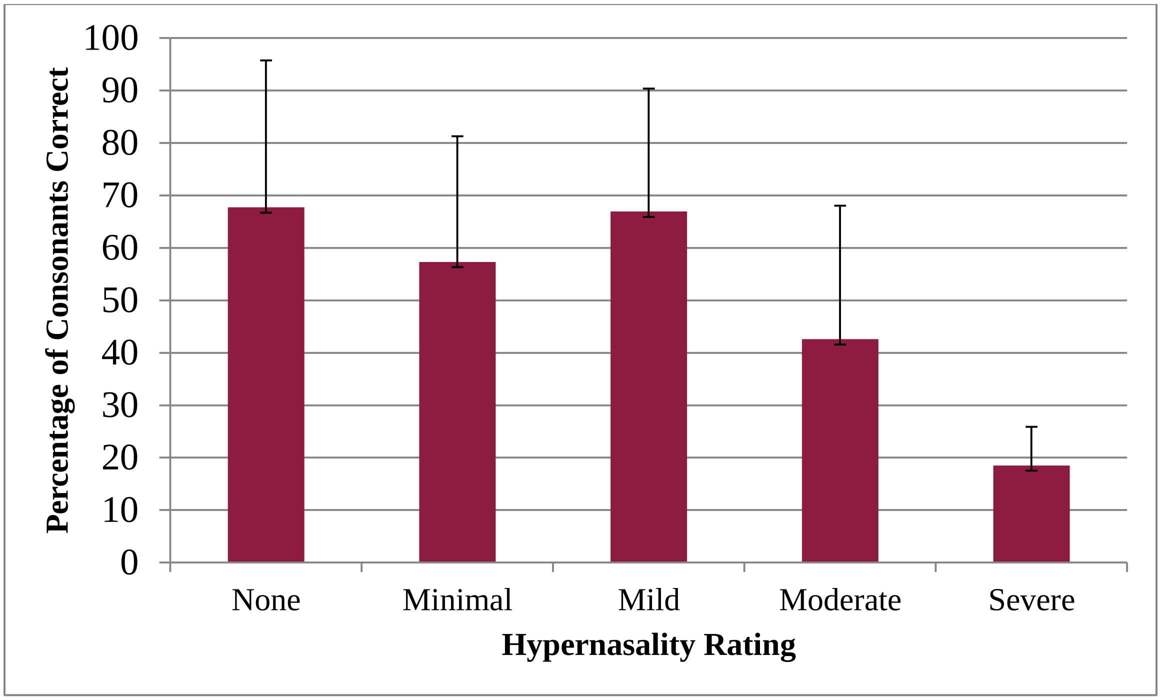
*Significant at $p<.01$

The strongest relationship was between fricative and affricate consonants, followed by fricative and stop consonants. Speech accuracy for fricatives is strongly correlated with production of both stops and affricates. These results suggest that fricative phonemes are the only manner class with significant associations between other high-pressure phoneme classes as well as the later-developing sound class of liquids.

Speech accuracy for fricatives differs by severity of hypernasality. Fricatives, which are later-acquired high-pressure phonemes, are particularly vulnerable to errors in the presence of velopharyngeal insufficiency. Therefore a one-way ANOVA was conducted to determine if speech accuracy for production of fricatives was different according to severity of hypernasality. Ratings of hypernasality included participants with no hypernasality (n=10), minimal hypernasality (n=21), mild hypernasality (n=14), moderate hypernasality (n=8), and severe hypernasality (n=3). There was a single outlier as assessed by boxplot, which included a participant with normal resonance who had significantly reduced speech accuracy for fricatives secondary to frequent use of phonological errors.

Data was normally distributed for each group, as assessed by Shapiro-Wilk test ($p > .05$); and there was homogeneity of variances, as assessed by Levene's test of homogeneity of variances ($p = .563$). Data is presented as mean ± standard deviation. Percentage of Consonants Correct (PCC) for fricatives was statistically significantly different between different ratings of hypernasality, $F(4, 51) = 3.623$, $p < .05$. Speech accuracy for fricatives decreased from the group with no hypernasality (67.71 ± 28.04), to mild hypernasality (66.92 ± 23.46), to minimal hypernasality (57.33 ± 23.96) to moderate hypernasality (42.58 ± 25.48) to severe hypernasality (18.51 ± 7.35) resonance groups, in that order. Tukey post hoc analysis revealed that the decrease from no hypernasality to severe hypernasality (49.20) was statistically significant (*p*=.028), as well as the decrease from mild hypernasality to severe (48.41, *p*= .024), but no other group differences were statistically significant. Figure 1 demonstrates how speech accuracy for fricatives differs according to hypernasality ratings.

Figure 1. Speech accuracy for fricatives according to severity of hypernasality.



Speech Accuracy by Place of Articulation

Children with CP+/-L demonstrated differences in speech accuracy, measured by percentage of consonants correct, across different places of articulation. Descriptive statistics for different places of speech articulation are reported in table 10. Children with CP+/-L demonstrated highest speech accuracy for bilabial place of articulation, followed by velar place of articulation. There was a substantial decrease for speech accuracy for alveolar place of production, with palatal place of production having the lowest speech accuracy. The variability in speech accuracy was greatest for palatal place of articulation.

Table 10. Descriptive statistics for speech accuracy by place of consonant production.

Place of Production	Mean (S.D.)
Bilabial Place of Production	93.39 (9.72)
Alveolar Place of Production	66.61 (23.57)
Palatal Place of Production	46.01 (35.98)
Velar Place of Production	87.88 (18.81)

Descriptive statistics by age group for speech accuracy by place of consonant production is presented in Table 11. Variability for speech accuracy by place of production was large across the age range of the study.

Table 11. Descriptive statistics for place of articulation by age group.

Place of Production	4-year-old (*n*=22) Mean (S.D.)	5-year-old (*n*=16) Mean (S.D.)	6-year-old (*n*=10) Mean (S.D.)	7-year-old (*n*=8) Mean (S.D.)
Bilabials	91.20 (11.12)	93.52 (11.07)	96.36 (4.69)	95.45 (6.87)
Alveolars	67.63 (20.03)	64.58 (22.36)	71.44 (23.83)	61.83 (35.88)
Palatals	36.77 (36.32)	60.49 (31.65)	38.75 (40.16)	51.56 (33.70)
Velars	88.64 (18.86)	90.40 (10.10)	81.25 (23.75)	89.06 (26.25)

The relationships between different places of articulation were examined. Visual inspection of scatterplots with linear best fit lines as well as with smoothed loess lines were used to establish whether linear relationships were present. Correlation analysis was only conducted for the relationships that were linear, including the following pairs of place classes: (a) bilabial with velar phonemes, (b) alveolar with palatal phonemes, (c), alveolar with velar phonemes, and (d) palatal with velar phonemes. The Pearson product moment correlations are presented in table 12 with a summary of each relationship.

Table 12. Summary of relationships between pairings of places of articulation.

Pairings between different places	Pearson Product	Description of Relationship

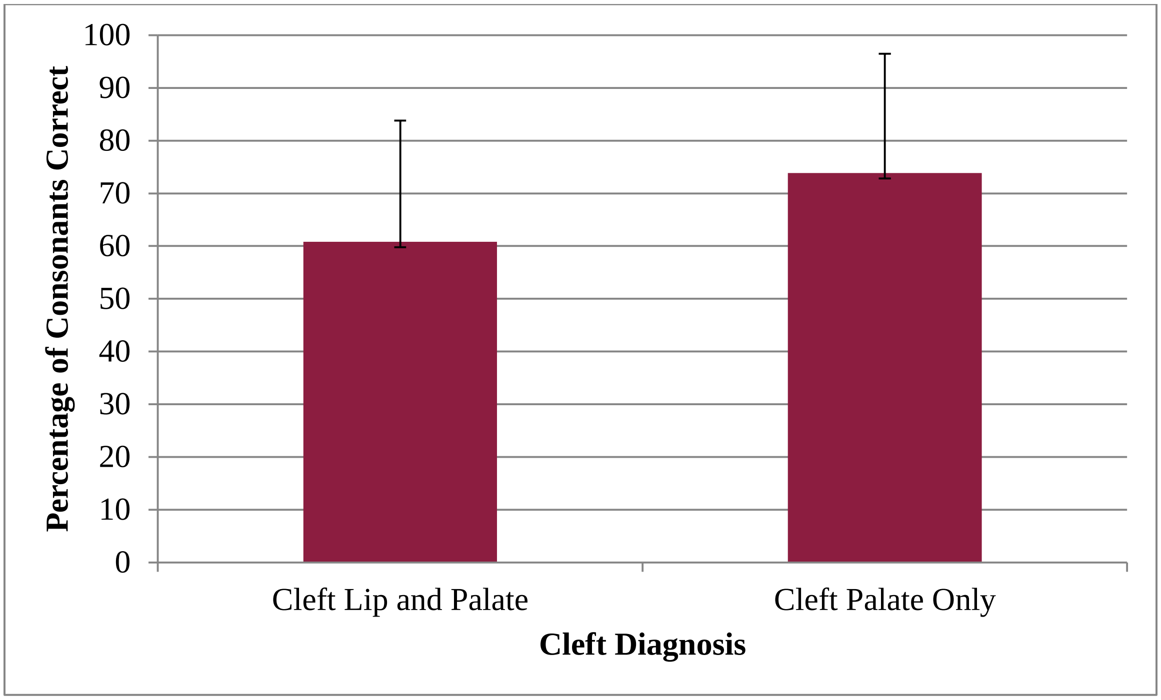
of articulation	Moment Correlation Statistic	
Bilabials with Velars	0.24	Nonsignificant relationship
Alveolars with Palatals	0.47*	Moderate positive correlation
Alveolars with Velars	0.50*	Strong positive correlation
Palatals with Velars	0.38*	Moderate positive correlation

*Significant at $p<.01$

The strongest relationships included that between alveolar place of production and velar place of production, as well as the relationship between alveolar place of production and palatal place of production. Bilabial consonants were not significantly correlated with any other place of articulation.

Speech accuracy for alveolar consonants differs by cleft type. A one-way ANOVA was conducted to determine if speech accuracy for production of alveolar consonants was different according to clefts with lip and alveolar involvement. Groups included individuals with cleft palate only (n=25) and individuals with cleft lip and palate (n=31). There were no outliers, and data was normally distributed for each group, based on visual inspection of Q-Q plots. There was homogeneity of variances, as assessed by Levene's test of homogeneity of variances (p= .682). Data is presented as mean ± standard deviation. Percentage of Consonants Correct (PCC) for alveolar consonants was statistically significantly different between different cleft groups, $F(4, 51) = 4.501$, $p<.05$. Speech accuracy for alveolar consonants decreased from individuals with cleft palate only (73.83 ± 22.63) to individuals with cleft of lip and palate (60.79 ± 23.02). Speech accuracy for alveolar consonants according to cleft type is presented in Figure 2.

Figure 2. Speech accuracy for alveolar consonants according to cleft diagnosis.



Speech Accuracy for Fricatives according to VPI, History of Secondary Surgery, and Age

A multiple regression was run to predict speech accuracy for fricative consonants from age, hypernasality, and history of secondary surgery. There was linearity as assessed by partial regression plots and a plot of studentized residuals against the predicted values. There was homoscedasticity, as assessed by visual inspection of a plot of studentized residuals versus unstandardized predicted values. There was no evidence of multicollinearity, as assessed by tolerance values greater than 0.1. There were no studentized deleted residuals greater than ±3 standard deviations, no leverage values greater than 0.2, and values for Cook's distance above 1. The assumption of normality was met, as assessed by a Q-Q Plot. The multiple regression model statistically significantly predicted percentage of consonants correct for fricative consonants, $F(3, 52) = 3.284$, $p < .05$, adjusted $R^2 = .11$. The only predictor variable that added significantly to the prediction was rating of hypernasality, $p < .05$. Regression coefficients and standard errors can be found in Table 13. In summary, severity of hypernasality was a significant predictor of PCC for fricative consonants, although age and history of secondary surgery did not contribute significant prediction of speech accuracy for fricative consonants.

Table 13. Summary of multiple regression analysis for fricative consonants.

Variable	Unstandardized regression coefficient	Standard error of the coefficient	Standardized Coefficient
Intercept	42.88	17.85	
Age	4.82	2.98	0.21
Rating of Hypernasality	-7.39*	3.09	-0.31
History of Secondary Surgery	-2.43	7.68	-0.4

*Significant at $p<.05$

Speech Accuracy for Total Consonants according to VPI, History of Secondary Surgery, and Age

A multiple regression was run to predict speech accuracy for total consonants from age, hypernasality, and history of secondary surgery. There was linearity as assessed by partial regression plots and a plot of studentized residuals against the predicted values. There was homoscedasticity, as assessed by visual inspection of a plot of studentized residuals versus unstandardized predicted values. There was no evidence of multicollinearity, as assessed by tolerance values greater than 0.1. There were no studentized deleted residuals greater than ±3 standard deviations, no leverage values greater than 0.2, and values for Cook's distance above 1. The assumption of normality was met, as assessed by a Q-Q Plot. The multiple regression model did not significantly predict percentage of consonants correct for total consonants, $F(3, 52) = 2.678$, $p=0.057$, adjusted $R^2 = .084$. In summary, these factors expected to be related to speech accuracy did not significantly contribute to prediction of speech accuracy for total consonants. When all consonant classes are considered, hypernasality was not a significant predictor of speech accuracy, even though it significantly predicted accuracy for fricative consonants.

Speech Error Usage: Aim Two Results

Speech accuracy is an important measure of the maturity of the child's speech system in relation to typical speech production; however, it does not provide information about the types of speech errors occurring. A detailed analysis of speech error type provides crucial information regarding the possible etiology of speech errors, including dental malocclusions, velopharyngeal insufficiency, developmental speech disorders, or a combination of these etiologies. Therefore, analyzing speech error types is crucial for understanding the nature of speech articulation disorders and what interventions are necessary for their remediation (e.g., secondary palate surgery to resolve VPI along with speech therapy to remediate use of compensatory articulation errors). This research question aimed to provide detailed analysis of speech error types across the categories of cleft-related articulation errors (i.e., anterior oral articulation errors, posterior articulation errors, non-oral compensatory articulation errors, passive cleft speech characteristics) and developmental/phonological errors (as outlined by Khan-Lewis Phonological Assessment). Additional error categories included ingressive speech errors and phonological nasal substitutions.

The primary speech errors produced by children in this sample included anterior oral cleft speech characteristics (CSCs), compensatory articulation errors, and developmental/phonological speech errors. Developmental/phonological errors were produced most frequently by children with CP+/-L in this sample. Lower frequency error types included posterior CSCs, passive CSCs, ingressive speech errors, and phonological nasal substitutions. Means and standard deviations for each error category along with total speech errors produced during AESS sentences are reported in Table 14.

Table 14. Descriptive statistics for speech error categories and total speech errors.

Error Type	Mean (S.D.)
Anterior Cleft Speech Characteristics	3.98 (3.61)
Posterior Cleft Speech Characteristics	0.48 (1.81)
Compensatory Articulation Errors	3.11 (5.16)
Passive Cleft Speech Characteristics	0.43 (1.78)
Developmental/Phonological Errors	9.07 (5.67)
Ingressive Errors	0.41 (2.03)
Phonological Nasal Substitutions	0.30 (1.04)
Total Speech Errors	17.79 (10.23)

Analysis of speech error types across age yields information on whether types of speech errors are consistent across this sample of four to seven-year-old children. Age as a factor that influences speech production is explored in more detail below. Descriptive statistics for each error category are presented according to age in Table 15.

Table 15. Speech error category descriptive statistics by age group.

Error Category	4-year-old (*n*=22) Mean (S.D.)	5-year-old (*n*=16) Mean (S.D.)	6-year-old (*n*=10) Mean (S.D.)	7-year-old (*n*=8) Mean (S.D.)
Anterior Oral CSCs	3.77 (3.84)	4.63 (2.53)	2.80 (3.62)	4.75 (4.86)
Posterior Oral CSCs	0.14 (0.35)	0.19 (0.40)	1.00 (2.49)	1.38 (3.89)
Compensatory Errors	3.18 (4.89)	2.56 (3.46)	2.20 (3.52)	5.13 (9.46)
Passive CSCs	0.59 (2.56)	0.19 (0.54)	0.30 (0.95)	0.63 (1.77)
Developmental Errors	11.32 (5.98)	8.81 (4.98)	8.40 (5.21)	4.25 (3.77)
Ingressive Errors	0.36 (1.71)	0.13 (0.34)	1.30 (4.11)	0 (0)

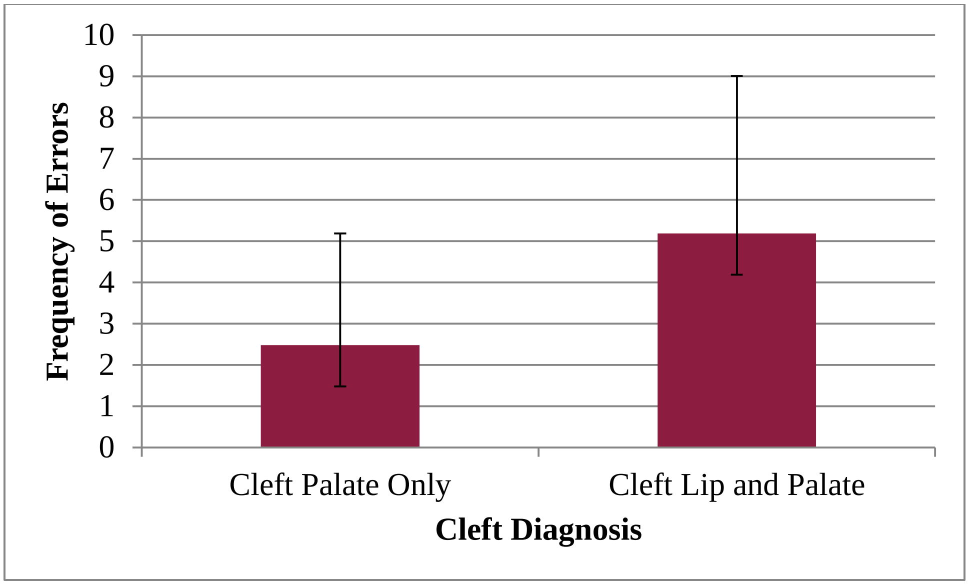
Nasal Substitutions	0.45 (1.50)	0.13 (0.34)	0.40 (0.97)	0.13 (0.35)
Total Errors	19.82 (10.10)	16.63 (7.53)	16.40 (11.36)	16.25 (14.41)

The following factors were explored as important characteristics that may impact rate of speech error usage: type of cleft palate (cleft palate only or cleft lip and palate), severity of hypernasality, history of secondary palate surgery, and age. Given the number of statistical tests being conducted, a Bonferroni correction was applied to reduce the rate of Type I error. The Bonferroni correction set the alpha level at 0.005.

Cleft Type and its Effect on Speech Error Rates

Cleft type in terms of cleft lip and palate versus cleft palate only was examined for its effect on anterior cleft speech characteristics. Number of anterior CSCs was statistically significantly different between different cleft types, $F(1, 54) = 8.98$, $p < .005$. Children with cleft lip and palate used significantly more anterior CSCs than children with cleft palate only. Figure 3 presents frequency of anterior CSCs according to cleft diagnosis. The effect of cleft type was nonsignificant for use of compensatory articulation errors, $F(1, 54) = 1.163$, p=0.286, and for use of developmental/phonological errors, $F(1, 54) = 0.212$, p=0.647.

Figure 3. Frequency of anterior oral CSCs according to cleft diagnosis.



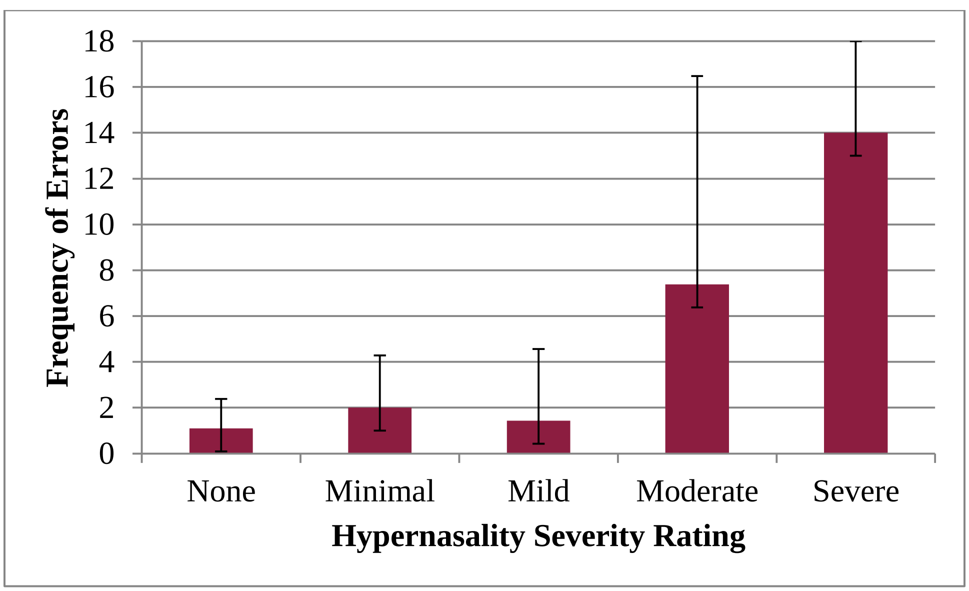
Severity of Hypernasality and its Effect on Speech Error Rates

The effect of hypernasality was examined for production of compensatory errors and production of passive cleft speech characteristics, which are consonant production differences occurring secondary to velopharyngeal insufficiency. Number of compensatory articulation errors was statistically significantly different between different ratings of hypernasality, $F(4, 51) = 9.020$, $p < .005$. Number of compensatory articulation errors was comparable for the hypernasality ratings of no hypernasality, minimal hypernasality, and mild hypernasality. Use of compensatory errors was much higher for individuals with moderate hypernasality, and was the highest for individuals with severe hypernasality. Descriptive statistics for frequency of compensatory error usage according to hypernasality rating is presented in Table 16. Tukey post hoc analysis revealed use of compensatory errors was significantly higher for individuals with moderate hypernasality compared to individuals with normal resonance (+6.28), individuals with minimal hypernasality (+5.38), and for individuals with mild hypernasality (+5.95). Additionally, use of compensatory errors was significantly higher for individuals with severe hypernasality compared to individuals with normal resonance (+12.90), individuals with minimal hypernasality (+12.00), and for individuals with mild hypernasality (+12.57). Use of compensatory articulation errors was not significantly different for individuals with moderate versus severe hypernasality. Figure 4 displays frequency of compensatory use according to hypernasality rating.

Table 16. Descriptive statistics for compensatory error usage by hypernasality rating.

Severity of Hypernasality	Mean (S.D.)
None	1.10 (1.29)
Minimal	2.00 (2.28)
Mild	1.43 (3.31)
Moderate	7.38 (9.10)
Severe	14.00 (4.00)

Figure 4. Frequency of compensatory error use by hypernasality rating



Number of passive cleft speech characteristics was statistically significantly different between different ratings of hypernasality, $F(4, 51) = 11.20$, $p < .005$. Use of passive cleft speech characteristics was highest for individuals with severe hypernasality, which is in line with expectations. Use of passive CSCs was not present or rarely present in speech production of individuals with none, minimal, mild, or moderate hypernasality. Tukey post hoc analysis revealed the use of passive CSCs increased significantly between moderate hypernasality and severe hypernasality (+4.46).

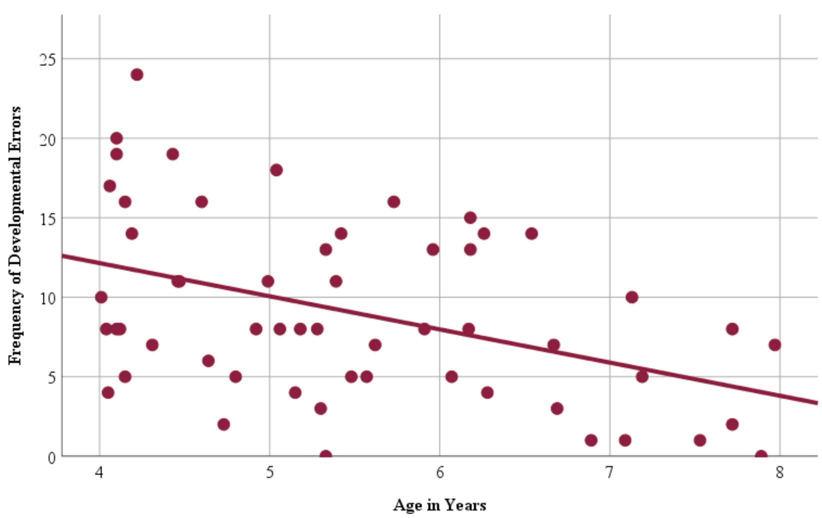
History of Secondary Surgery and its Effect on Speech Error Rates

Number of compensatory articulation errors was not statistically significantly different for children with history of secondary palate surgery compared to children with no secondary palate surgery, $F(1, 54) = 0.106$, $p=0.746$. Children who had received secondary surgery for palate function did not use significantly fewer compensatory articulation errors than children with no secondary surgery for speech. Even though some children had received prior velopharyngeal management, compensatory articulations are habituated errors that often require speech therapy to remediate. To further explore the relationship between history of velopharyngeal surgery and speech accuracy, additional information regarding type of secondary palate surgery, age at secondary palate surgery, speech outcome after surgery, and history of appropriate speech therapy goals would be additional characteristics to investigate.

The Effect of Age on Speech Error Rates

Multiple regression analysis was run to predict rate of speech error usage according to age, since age was measured on a continuous scale. Three multiple regression analyses were conducted to examine the effect of age on the three primary speech error types. Age did not significantly predict number of speech errors for anterior CSCs, $F(1, 54) = 0.47$, p=0.8,3 or for compensatory articulation errors, $F(1, 54) = 0.186$, p=0.67. Rate of anterior CSC usage and compensatory usage did not change as children aged. However, age was a significant predictor of use of developmental/phonological speech errors. A linear regression established that age significantly predicted number of phonological errors produced during the American English Sentence Sample production, $F(1, 54) = 12.20$, $p<.01$, accounting for 18% of the variation in the number of developmental/phonological errors used, with an adjusted R-squared of 0.169. For each year the child aged, the number of developmental/phonological errors decreased by 2.09 errors. Figure 5 presents frequency of developmental/phonological error use according to age. These results are in line with expectations that as a child's speech articulation skills develop between age four and age seven, their use of developmental/phonological errors should decrease.

Figure 5. Frequency of developmental error use by age in years.



Speech Accuracy by Context of Production: Aim Three Results

Evaluating articulation skills using a single-word naming task is recognized as a of the gold standard assessment tool; however, it is also important to evaluate speech production in connected speech contexts. In order to understand the concurrent validity of speech accuracy during a sentence repetition task (i.e., AESS) with speech accuracy for single-word naming task (i.e., GFTA-3), the relationship between these speech measures was explored. Descriptive statistics for speech accuracy at the single-word level and at the sentence level are provided in Table 17. Mean speech accuracy during assessment of articulation at the single word level was slightly higher than mean speech accuracy during a sentence repetition task. The variability of speech accuracy, as measured by standard deviations, was similar across both contexts of speech production.

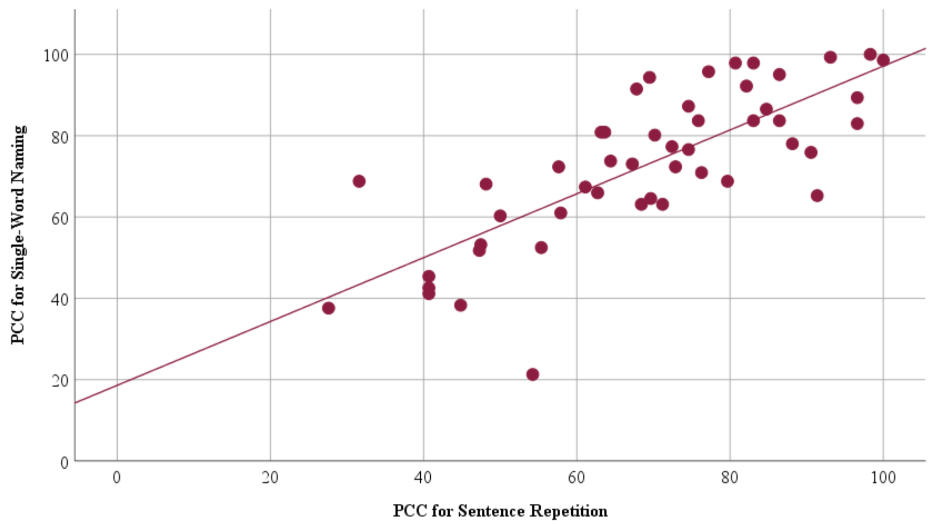
Table 17. Speech accuracy by context of production.

Speech Production Context	Mean (S.D.)
Single-words (GFTA-3)	72.89 (18.65)
Sentence Repetition (AESS)	69.14 (18.19)

A Pearson's product-moment correlation was conducted to assess the relationship between PCC for a single-word articulation assessment and PCC for the AESS. This assessment of speech accuracy was conducted to provide information on concurrent validity between speech accuracy for the GFTA-3 and the AESS. Preliminary analyses showed the relationship to be linear with both variables normally distributed, as assessed by Shapiro-Wilk's test ($p > .05$), and there were no outliers. There was a statistically significant, strong positive correlation between PCC for the GFTA-3 and PCC for the AESS, $r(54) = .77$, $p < .01$, with PCC for the GFTA-3 explaining 58% of the variation in

PCC for the AESS. Figure 6 demonstrates the relationship between PCC for the sentence repetition task and PCC for the single-word naming task. In summary, there is a strong relationship between speech accuracy on a single-word naming test and speech accuracy during a sentence repetition task.

Figure 6. Relationship between PCC for a sentence repetition task and a single-word naming task.



In order to make comparisons between the current sample and previous intercenter outcomes studies, the proportion of the sample with disordered speech articulation skills according to standard scores for the single-word naming test are presented. In the current sample, 65.3% of the participants (*n*=32) presented with a speech articulation disorder based on standardized assessment of speech articulation, while 34.7% (*n*=17) demonstrated speech articulation skills within the average range based on standardized testing.

CHAPTER 5

DISCUSSION

Speech Accuracy: Aim One Discussion

Total Speech Accuracy

The average speech accuracy for this sample of children with CP+/-L was 69.25, indicating reduced speech accuracy on average compared to age expectations. For children with typical speech development with no history of clefting, age expectations for PCC at 36 months is 87% (Stoel-Gammon & Williams, 2013). This sample of children with CP+/-L included an older age range than the normative data presented by Stoel-Gammon and Williams (2013); however, this sample, on average, demonstrated speech accuracy more than 17 points below the normative data available. When compared to the severity ratings for PCC developed by Shriberg and Kwiatkowski (1982), the average PCC demonstrated by this sample indicates a mild-moderate speech disorder.

Children in the current study presented with, on average, lower speech accuracy measured by PCC compared to other published research on speech accuracy in children with cleft palate. Many published studies report on the speech accuracy of 5-year-olds with cleft palate; therefore, comparisons between the current study and previous published reports of speech accuracy will be made for both the full sample and specifically for the 5-year-olds in the current sample (n=16).

For the Scandcleft Project Trial 3, Persson and colleagues (2020) reported the proportion of participants demonstrating age-appropriate PCC level during a single-word naming task for 136 five-year-olds with unilateral cleft lip and palate. In total, 38% of the participants demonstrated an age-appropriate PCC level above 90% accuracy. In the current study of children with CP+/-L between the ages of 4 and 7, only 12.5% of the sample reached speech accuracy greater than 90%. Additionally Persson and colleagues (2020) reported 38% of their sample of 5-year-olds with unilateral cleft lip and palate had PCC values less than 79. In the current sample, 69.6% of the sample demonstrated PCC less than 79. When evaluating the total speech accuracy of 5-year-olds with unilateral cleft lip and palate in the current sample (*n*=7), all seven of these participants demonstrated PCC values less than 79.

In a study of articulation proficiency of five-year-olds with cleft palate (Malmenholt et al., 2019), 39% of the sample demonstrated age-appropriate articulation proficiency on a single-word naming task, as indicated by PCC value of 91% or higher based on linguistic norms in Swedish. In the current project only two 5-year-old children (12.5% of the five-year-olds in the sample) demonstrated PCC values greater than 90%. In fact, 13 of the five-year-olds in the current study (81.3% of the five-year-olds) demonstrated PCC values lower than 79.

During a connected speech task (e.g., story retell task) completed by 29 5-year-olds with UCLP the median PCC was 89, with a range of 24 – 100 (Klinto et al., 2016). This is 20 points higher than the mean PCC of the current sample of 4 to 7-year-old children during connected speech production (i.e., sentence repetition task) and 18 points higher than the mean PCC of the 5-year-olds in the current sample (PCC range for five-year-olds: 47.27 – 96.61).

Speech Accuracy by Manner of Articulation

One aim of this study was to examine speech accuracy according to manner of articulation. Infants with CP+/-L have early delays in acquiring high-pressure phonemes secondary to unrepaired cleft palate. Children with persistent VPI may continue to have difficulty with producing these phonemes. In the current study speech accuracy was highest for nasal and glide consonants, which is in line with expectations given these sound classes are early-developing and are not restricted by VPI prior to initial palate repair. Following nasal and glide consonants, the manner class of stop consonants was produced with the highest speech accuracy. While stop consonants are high-pressure phonemes, these are the next earliest manner class to be acquired according to typical speech development. There was a large discrepancy in speech accuracy between stop consonants and fricative consonants, which is notable since stop consonants are earlier-acquired sounds than fricative consonants. This study found that fricative consonants were a particularly vulnerable manner class for children with CP+/-L to produce accurately. These later-developing high pressure phonemes may be impacted by compensatory articulation errors secondary to VPI, dental-occlusal related errors associated with clefting, or developmental errors. This study found that speech accuracy was lowest for affricate and liquid consonants, which are the latest acquired manner classes according to typical speech development. Despite the structural differences known to influence speech production in children with CP+/-L, this sample, on average, followed the trajectory expected by typical speech development for the order of manner classes to be produced correctly. It is noted that variability of speech accuracy was largest for fricative, affricate, and liquid consonants, indicating that children with CP+/-L demonstrated variable performance in articulating these later-acquired phoneme classes.

Few studies have reported speech accuracy according to manner of consonant production in children with CP+/-L between 4 and 7 years of age. Jorgenson and

Willadsen (2020) reported PCC for all obstruent consonants (i.e., high-pressure consonants) for a subset of five-year-old children included in the Scandcleft Project. Mean speech accuracy for obstruents was 84 (S.D. 17) in the report by Jorgenson and Willadsen (2020). Obstruents examined by Jorgenson and Willadsen included primarily stop consonants and only two fricative consonants. In the current study, mean speech accuracy for stops, fricatives, and affricates (each class of high-pressure phonemes) was 86.85, 57.39, and 46.34 respectively. Since the analysis by Jorgenson and Willadsen (2020) did not report speech accuracy for manner classes separately, it is not possible to know if there was also a large discrepancy between speech accuracy for stop versus fricative phonemes as was observed in the current project.

While previous research on speech accuracy according to specific manner classes is limited for children with CP+/-L ages 4 through 7 years, previous work has examined speech accuracy by manner of production for toddlers with cleft palate (Scherer et al., 2013). The speech accuracy for each manner class was much lower for toddlers in the study by Scherer and colleagues compared to the current sample; however, the order of highest speech accuracy to lowest speech accuracy was similar for both samples. Both the current sample and the toddler sample demonstrated highest speech accuracy for nasals and glides, followed by stop consonants. For both samples, there was a large discrepancy between speech accuracy for stop consonants and fricative consonants. Affricates and liquids were produced with the lowest accuracy across both samples. Interestingly, the pattern for speech accuracy across manners of articulation was consistent across both samples.

Relationships between Manner Classes

The relationships among speech accuracy for different manner classes were examined to understand which classes had the strongest correlation. The strongest relationships were between fricative and affricate consonants, followed by fricative and stop consonants. Speech accuracy for fricatives was strongly correlated with production of both stops and affricates. These results support that fricative phonemes serve as a bridge between the acquisition of stop phonemes and affricate phonemes, which support typical speech development expectations.

Speech Accuracy for Fricatives Differs by Severity of Hypernasality

Speech accuracy for fricative consonants differed significantly according to severity of hypernasality ratings. Individuals were severe hypernasality had the lowest speech accuracy, followed by individuals with moderate hypernasality. It is in line with expectations that individuals with greater severity of VPI would have lower speech accuracy for fricatives, which is the largest class of high pressure consonants.

Speech Accuracy by Place of Articulation

Speech accuracy differed according to place of articulation. Children with CP+/-L demonstrated highest speech accuracy for bilabial place of articulation, followed by velar place of articulation. There was a substantial decrease for speech accuracy for alveolar place of production, with palatal place of production having the lowest speech accuracy. These findings are in line with expectations that children with CP+/-L have greater difficulty with place of articulation for structures that have been affected by clefting. The variability in speech accuracy was greatest for palatal place of articulation.

Previous literature has rarely reported speech accuracy by place of articulation for children with CP+/-L aged 4 to 7. However, one study by Jorgenson and Willadsen

(2020) did describe speech accuracy for bilabial, alveolar, and velar phonemes for 5-year-old Danish children. Their sample of 108 5-year-olds produced bilabial phonemes with 91% accuracy (S.D. = 0.18), which is comparable to the current study's findings for bilabial speech accuracy (Mean=93.39). The sample by Jorgenson and Willadsen produced alveolar phonemes with 78% (S.D. = 25) accuracy, which was higher than the mean from the current study (66.61). Finally, the Danish children produced velar consonants with 85% accuracy (S.D. = 28), which is similar to the current study's findings for velar place of articulation, with a mean of 87.88. Data on palatal place of articulation was not reported by Jorgenson and Willadsen. Both the current study as well as the study by Jorgenson and Willadsen (2020) found that speech accuracy was higher for bilabial and velar place of articulation, with a substantial decrease in accuracy for alveolar phonemes.

Relationships between Place of Articulation

The strongest relationship between different places of articulation was for alveolar and palatal consonants. This finding is interesting given that alveolar and palatal articulation would both be significantly impacted by presence of a cleft. While velar placement would also be impacted by cleft of the soft palate, children with cleft typically acquire phonemes with posterior placement earlier.

Speech Accuracy for Alveolar Consonants Differs by Cleft Type

Children with cleft lip and palate demonstrated significantly lower speech accuracy for alveolar targets than children with clefts involving the secondary palate only. Since few studies have reported speech accuracy for alveolar consonants separately from other manner classes, it is not possible to identify if this finding is supported by previous literature.

Speech Error Usage: Aim Two Discussion

The primary speech errors produced by children in this sample included developmental/phonological errors, anterior oral cleft speech characteristics (CSCs), and compensatory articulation errors. Posterior oral CSCs, passive CSCs, ingressive errors, and phonological nasal substitutions occurred infrequently in the current sample.

Cleft Speech Characteristics

A national audit conducted by the United Kingdom between 2001 and 2003 revealed that 60% of its sample of 1,110 children with CP+/-L at age five presented with no cleft-related articulation difficulties requiring speech therapy (Britton et al., 2014). This standard was operationalized as no cleft speech characteristics other than dentalization and less than two consonants affected by lateralization, palatalization, or double articulation. This audit focused specifically on cleft-related articulation errors included in the CAPS-A protocol and did not take developmental/phonological errors into account.

In order to make comparisons to the results reported by Britton and colleagues (2014) the percentage of participants with no cleft-related speech characteristics according to CAPS-A categories was calculated. In the current sample, 23.2% of participants demonstrated no anterior oral cleft speech characteristics. This speech error category included dentalization errors that were deemed acceptable for 5-year-old children to produce in the results reported by Britton and colleagues. There were 82.1% of participants in the current sample with no posterior oral cleft speech characteristics. Only 32.1% of the sample demonstrated no occurrences of non-oral compensatory articulation errors. Further review of use of compensatory errors in the current sample revealed that 37.5% of the sample used only 1 or 2 compensatory errors throughout the speech sample, while 30.4% of the sample used 3 or more compensatory errors during the speech sample. For passive cleft speech characteristics, 89.3% demonstrated no errors in this category. The current study of 4-year-old to 7-year-old children had more participants using anterior oral CSCs (including dentalization errors) than the data compared to the benchmarks provided by Britton et al. (2014). A larger percentage of participants from the current study used compensatory errors, although many children (37.5% of the sample) only used 1 or 2 compensatory errors across the sentence repetition task. The benchmarks described by Britton and colleagues provide useful comparisons for other studies examining use of cleft speech characteristics.

The Scandcleft Project reports speech articulation outcomes for 5-year-olds with unilateral cleft lip and palate who participated in the randomized controlled trials that assessed surgical protocols (Hammarstrom et al., 2020; Persson et al., 2020; Willadsen et al., 2019). The project included a series of three trials comparing a common surgical procedure (i.e., lip and soft palate closure at three to four months; hard palate closure with vomer flap at 12 months) to three local procedures, including the following: a) delayed hard palate closure at 36 months of age; b) single-stage palate closure protocol with lip closure at 3-4 months and closure of hard and soft palate at 12 months; c) lip closure at 3-4 months combined with a single-layer closure of the hard palate using vomer flap, followed by soft palate closure at 12 months. These local surgical procedures are compared to the common procedure in Scandcleft Trials 1, 2, and 3 respectively. The Scandcleft speech outcomes described below were assessed using a single-word naming test with phonemes that occurred across the five relevant languages (i.e., Danish, English, Finnish, Norwegian, Swedish). The project reported active cleft speech characteristics (passive cleft speech characteristics occurring secondary to VPI were not included in analyses), and sub-divided its active CSCs into oral versus non-oral CSCs. Scandcleft defined oral CSCs as retracted articulations occurring in the oral cavity, including palatal, velar, or uvular placement. Non-oral CSCs include glottal and pharyngeal productions, active nasal fricatives, and nasal substitutions for unvoiced plosives and fricatives. These error sub-categories do not directly correspond to the cleft speech error categories used by the CAPS-A which includes anterior oral CSCs, posterior oral CSCs, and non-oral compensatory CSCs. Proportion of participants using three or more speech errors per category according to study sample are presented in Table 18. This dissertation project presented data for five-year-old children with unilateral cleft lip and palate to match the Scandcleft participants, as well as data for all five-year-old children in the current study and the full sample of participants.

Table 18. Percentage of participants demonstrating three or more errors per category.

Study Sample	Active CSCs[a]	Anterior Oral CSCs	Posterior Oral CSCs	Non-Oral Compensatory Errors
Scandcleft Trial 1	Data not reported	32.9%[b]	32.9%[b]	13.9%[b] used non-oral compensatory errors; 6.3%[b] used glottal stops[c]
Scandcleft Trial 2	21.5% of all Trial 2 participants	Data not reported	Data not reported	Data not reported
Scandcleft Trial 3	47% of all Trial 3 participants	29% had "oral-retracted" CSCs, which corresponds to		28% of all Trial 3 participants

		both anterior and posterior CSCs for CAPS-A error categories		
Current Study: 5-year-olds with UCLP *n*=7	100%	85.7%	0%	42.9%
Current Study: 5-year-olds only *n*=16	81.25%	75%	0%	31.25%
Current Study: Full Sample *n*=56	75%	55.3%	3.6%	30.4%

a. For this dissertation, Active CSCs represents the sum of the other three error categories studied in this project, including a) anterior oral CSCs, b) posterior oral CSCs, and c) non-oral compensatory errors.
b. Percentage calculated by the first author for all of Scandcleft Trial 1 participants as reported in Willadsen et al., 2017, Table 10.
c. Willadsen et al., 2017 reported percentage of children using 3 or more glottal stops separately from children using 3 or more non-oral compensatory errors

Overall there was a larger proportion of children in the current sample demonstrating use of active cleft speech characteristics compared to the data available for Scandcleft Trials 2 and 3 (Hammarstrom et al., 2020; Persson et al., 2020). This substantial difference is likely related to the speech sample material being a sentence repetition task for the current study, while the Scandcleft project used a single word naming task. Differences in speech material are known to influence speech production skills (Klinto et al., 2011). Interestingly, the percentage of participants using non-oral compensatory errors was comparable for the current sample compared to Scandcleft Trial 3.

Developmental Speech Errors

The Scandcleft Trials also reported proportion of participants with developmental speech characteristics (Willadsen et al., 2017). Since few studies report developmental speech characteristics separately from overall speech proficiency in children with cleft palate, the data from the Scandcleft Trials are presented below. However, cross-linguistic differences do not allow direct comparisons between developmental/phonological speech characteristics in English with the errors considered developmental in nature for the Scandinavian languages used for assessment in Scandcleft. The Scandcleft study only coded for 7 different developmental errors relevant for the Scandinavian language of interest, including: a) errors in voicing, b) errors in aspiration, c) fronting, d) stopping, e) frication, f) interchangeable u se of fricatives, and g) interchangeable use of /n/ and /l/. The current study coded for 12 different developmental errors. Data for percentage of participants who used 3 or more developmental speech errors is reported in Table 19. For the participants in the current sample, it was age-appropriate for the children to be using developmental errors for later-developing sounds (e.g., /r/, th). Unfortunately the research on developmental speech errors for children with CP+/-L between the ages of four and seven is limited; therefore, comparisons to other English-speaking samples is not possible at this time.

Table 19. Percentage of participants demonstrating three or more developmental speech errors.

Study Sample	Developmental Speech Errors
Scandcleft Trial 1	Data not reported
Scandcleft Trial 2	44.1% of Trial 2 Participants
Scandcleft Trial 3	40% of Trial 3 participants
Current Study: 5-year-olds with UCLP (*n*=7)	100% of sample
Current Study: 5-year-olds only (*n*=16)	93.7% of sample
Current Study: 4-7 year-olds (*n*=56)	87.5% of study

Factors that Affect Speech Error Usage

Individual and treatment characteristics affected frequency of speech error usage. Specifically, presence of cleft lip was related to higher frequency of anterior oral CSCs. Severity of hypernasality influenced frequency of compensatory errors and passive CSCs. History of secondary surgery was not related to frequency of compensatory errors. Age significantly predicted developmental/phonological errors, but it did not predict frequency of other error categories (i.e., anterior oral CSCs or compensatory errors).

Previous research has not examined the impact of cleft type specifically on frequency of error usage, rather previous research has typically focused on impact of cleft type on overall consonant proficiency (Britton et al., 2014; Lohmander & Persson, 2008; Malmenholt, et al., 2019). This study contributes information on how clefting involving the lip, alveolus and palate may result in more frequent usage of anterior oral CSCs.

Substantial research has been completed to determine if staging of primary palate repair and timing of hard palate closure impact use of speech errors (Willadsen et al., 2017). However, history of secondary surgery as a variable that impacts frequency of compensatory error usage has not been specifically examined. This study revealed that compensatory error usage was not different for children with a history of velopharyngeal surgery versus children with no previous velopharyngeal management. Further research regarding type and timing of velopharyngeal management, along with history of speech therapy and content of speech therapy targets may be additional characteristics that influence use of compensatory errors.

Children demonstrating moderate or severe hypernasality used significantly more compensatory speech errors than children with normal, minimal, or mild hypernasality. For those children with moderate or severe hypernasality, additional velopharyngeal management to address VPI concerns along with quality speech therapy to remediate use of compensatory articulation errors is indicated based on results of this study. Finally, age was a significant predictor of developmental/phonological errors, indicating that older children in this sample used fewer developmental errors. Since children between the ages of 4 and 7 are continuing to acquire later-developing phonemes, this finding is in line with speech development expectations.

Understanding how these individual and treatment characteristics related to speech error usage is important for assessment practices and management decisions. Speech assessment should account for these different error categories when describing speech results. Different speech errors may require different speech therapy approaches (e.g., phonological intervention for developmental/phonological errors versus phonetic intervention for establishing accurate place of production for compensatory errors or anterior oral CSCs).

Speech Accuracy by Context of Production: Aim Three Discussion

Children in the current sample demonstrated slightly higher speech accuracy during a single-word naming task (i.e., GFTA-3 assessment) compared to sentence repetition of the AESS, although the mean speech accuracy level was similar. The variability was also similar for both methods of speech assessment. This finding is in contrast to expectations that children with CP+/-L would demonstrate higher speech accuracy skills during a single-word naming task compared to connected speech production. The nature of the sentence repetition task (i.e., a clinician modeling the target sentence) may have supported speech production skills for this connected speech context.

The significant strong positive correlation between speech accuracy for the GFTA-3 compared to speech accuracy for the AESS demonstrates concurrent validity for the AESS. The AESS, which samples speech according to the universal speech parameters (Henningsson et al., 2008) has validity for assessing speech production in children with cleft palate when compared to the GFTA-3, which has strong psychometric properties (Goldman & Fristoe, 2015).

In the current study, there was a high proportion of children presenting with speech disorders (65.3%). In the Americleft study, 68% of children demonstrated articulation errors atypical for their age, while 32% had normal speech articulation on the GFTA-2 (Chapman et al., 2017). Even though the Americleft speech study included 5-year-old and 6-year-old children with unilateral cleft lip and palate, its speech articulation outcomes according to standardized assessment of single word production was comparable to the current study of four to seven-year-old children with CP+/-L. A different research report indicated 61% of 5-year-old Swedish speakers had articulation proficiency below the average range, as measured by PCC more than 1 standard deviation below the Swedish normative values (Malmenholt et al., 2019).

Previous investigations of speech material for children with CP+/-L have revealed higher speech accuracy measured by PCC for word naming compared to sentence repetition tasks. Specifically, Klinto and colleagues (2011) reported median PCC of 86.4 (range: 38.5 – 100) for a word naming task, compared to median PCC of 81.5 (range: 18.8 – 99.0) for sentence repetition task in Swedish. The current study demonstrated a median PCC of 73.76 (range: 21 – 100) for the single word naming task (i.e., GFTA-3) and a median PCC of 70.68 (range: 27.59 – 100) for the sentence repetition task. While the median scores reported by Klinto and colleagues for the word naming task and the sentence repetition task were more than 10 points higher than the scores for the current study, there was a similar difference between the two speech tasks.

In summary, the current study demonstrated slightly higher speech accuracy for single word production compared to sentence repetition, although previous research has demonstrated a similar trend. Both single word naming tasks and sentence repetition tasks provide valuable information to a speech evaluation and should be included as components of a standard assessment protocol for children with CP+/-L.

Clinical Implications

This study has contributions to support speech assessment practices for children with CP+/-L between the ages of four and seven. Most importantly, using the AESS sentence repetition task to evaluate speech accuracy is strongly correlated with using the GFTA-3 single word naming assessment to determine speech accuracy. Given the strong psychometric properties of the GFTA-3, this is promising evidence to support use of the AESS sentence repetition task. While the goal of assessing single-word production is to evaluate a child’s best attempt at speech production, using a sentence repetition task is crucial for assessing performance in connected speech contexts. Connected speech production is an opportunity to evaluate speech skills as demonstrated by a child in more naturalistic contexts (e.g., conversation). The AESS isolates target phonemes so the impact of speech characteristics common in children with CP+/-L (e.g., hypernasality, nasal emission) can be evaluated using a valid speech sample. While the AESS has been used for the Americleft Speech project and has been adopted for uses in cleft palate clinics, its relationship to speech production skills in other contexts of production (e.g., single-word naming) has not yet been established prior to this study.

This study also supports assessing speech accuracy by manner and place of articulation in order to obtain detailed information about the strengths and areas of need in a child's developing speech sound system. Speech accuracy results by manner and place of articulation may support identifying specific phonemes or classes of phonemes to target in speech-language intervention. Evidence that speech accuracy for fricative phonemes is influenced by severity of hypernasality also provides clinicians with guidance to conduct a detailed assessment of those phonemes in children diagnosed with velopharyngeal insufficiency. Reduced speech accuracy for alveolar phonemes in individuals with clefts involving the lip and alveolus also provides guidance for clinicians to closely evaluate those phonemes in children with that diagnosis.

In addition to evaluating speech accuracy, conducting a detailed analysis of speech errors is beneficial for differential diagnosis of speech articulation and/or resonance disorders. This study examined both cleft speech characteristics included in the CAPS-A-AM protocol (i.e., anterior oral CSCs, posterior oral CSCs, non-oral compensatory errors, and passive CSCs) as well as developmental/phonological errors included as core processes of the Khan-Lewis Phonological Assessment. It is important for clinicians who evaluate speech production in individuals with CP+/-L to examine the type(s) of speech errors occurring in order to make appropriate management recommendations. Speech-language intervention approaches should also be selected based on types of speech errors present in a child's sample.

Of the different speech error categories evaluated in this study, age was only a significant predictor of frequency of developmental/phonological errors. Older children in this sample used significantly fewer developmental/phonological errors. This finding indicates that children with CP+/-L between the ages of four and seven still demonstrate developing phonological systems, which has implications for timing of speech assessments for this age range.

Children with clefts involving the lip and alveolus demonstrated significantly more anterior oral CSCs in this study. Since children with CP+/-L demonstrate transitional dentition and are anticipating or receiving alveolar bone grafting within this age range, it is important for the speech-language pathologist to collaborate with the cleft team orthodontist regarding the child's use of anterior oral CSCs. Understanding the child's dentition and the role it has in use of anterior oral CSCs will support the speech-language pathologist's assessment and associated recommendations.

Unsurprisingly, children with moderate and severe hypernasality used significantly more compensatory articulation errors than children with mild, minimal, or no hypernasality. This finding guides clinicians to recommend velopharyngeal management to address VPI as well as to recommend speech therapy focused on remediating use of compensatory articulation errors. Children with severe hypernasality used significantly more passive CSCs than any other hypernasality rating group, which also suggests that velopharyngeal management for severe hypernasality is necessary to support overall speech accuracy.

In summary, this study provides support for using the AESS sentence repetition task to evaluate speech articulation children with CP+/-L as well as suggestions for how to analyze speech production skills according to accuracy and use of errors. Characteristics that influenced speech production skills in children with CP+/-L aged four through seven should also be considered in clinical decision-making.

Limitations

This dissertation used a clinical registry from the Barrow Cleft & Craniofacial Center as its data source for speech samples versus recruiting participants for prospective data collection. While the use of the clinical registry yielded a larger sample size than would have been collected using prospective methodology, it is possible that sampling bias exists. The goal of BCCC is to conduct a speech-language evaluation for any child with a cleft palate followed by the BCCC cleft team between ages 4;0 and 5;6. Ideally, this cleft team timeline would mean that all children with CP+/-L receive a speech-language evaluation regardless of whether speech articulation or velopharyngeal insufficiency concerns exist. However, it is possible that children with CP+/-L without speech concerns do not attend the speech-language evaluation recommended by the cleft team timeline and do not have a speech-language evaluation documented in the clinical registry. Therefore, this sample may include a higher proportion of children with speech articulation concerns since those returning to the clinic may be more likely to have active speech concerns.

Initially this dissertation aimed to examine speech for children with CP+/-L between the ages of 3;0 through 7;11 since this is an important period for the development of speech articulation skills. As mentioned in the method section, only four

3-year-old children were able to complete the assessment protocol using the AESS sentence repetition task. Therefore the age range was adjusted to 4;0 through 7;11. For the eligible participants within this age range, the earliest speech assessment that contained production of the AESS was included in the project. This methodological decision of using the earliest available speech sample resulted in having more 4-year-old (*n*=22) and 5-year-old (*n*=16) participants compared to 6-year-old (*n*=10) and 7-year-old (*n*=8) participants. Any investigation of age in this project was conducted using age as a continuous variable across the range of 4;0 through 7;11 to reduce any impact of unequal sample size by age group on the results. Additionally, using age as a continuous variable follows statistical guidelines to avoid dichotomizing continuous variables (Altman & Royston, 2006). Future research may aim to recruit more 6-year-old and 7-year-old participants to have greater representation of speech skills at these ages.

Phonetic transcription and ratings of perceptual parameters of speech were performed on audio-recorded speech samples. The procedures used for audio-recording are standardized in order to yield a high quality audio recording of the clinical speech sample as a part of the standard of care at BCCC. Any audio files that included technical issues or significant environmental noise were excluded from the project. Despite these measures to ensure high fidelity speech sample recordings, audio-recordings do not allow the listener to visually confirm the perceptual analysis of speech, especially in terms of speech articulatory placement. Future studies of speech production for children with CP+/-L should include video and audio-recordings of speech for this reason.

Inter-rater reliability for phonetic transcription was conducted based on speech samples that were re-transcribed by a trained undergraduate student. The student demonstrated strong knowledge of cleft-related articulation errors and was skilled in phonetic transcription; however, the student had limited experience transcribing cleft-related speech errors. For future studies, a speech-language pathologist with sufficient experience and expertise in the perceptual assessment of cleft palate speech should conduct phonetic transcription for reliability analysis.

The research question regarding performance on the single-word naming test (GFTA-3) was addressed in order to anchor the results of the current sample to data that has been reported by larger inter-center outcomes studies, including the Americleft Speech Study. Additionally, the GFTA-3 is routinely used in the clinical evaluation of speech production to make therapy recommendations, so it is important to understand how the speech results for the sentence repetition task in the current study compared to the single-word naming task. However, the data for the GFTA-3 for the current study was reported from the clinical registry as a part of the speech-language evaluation documentation. The GFTA-3 is not audio-recorded at BCCC, so it was not possible to conduct phonetic transcription as a primary rater or to perform reliability on these results. The GFTA-3 results were scored and analyzed by three different speech-language pathologists with expertise in assessing the speech skills of children with cleft palate, but these data were clinical in nature. Additionally, there were seven participants who did not have GFTA-3 data available on the same day they completed the sentence repetition task, which meant the sample size was smaller for the research question focused on the GFTA-3.

Despite these limitations, the current study adds valuable information regarding speech accuracy and speech error usage to the body of literature on speech articulation skills of individuals with cleft palate.

Future Directions

This cross-sectional study has added valuable information about speech accuracy and speech error usage in children with CP+/-L between the ages of 4 and 7 to our field of study. In the current study the characteristics of age and history of secondary palate surgery did not have a significant impact on speech accuracy. Severity of hypernasality significantly predicted speech accuracy for fricative phonemes, but not for total speech accuracy. For speech errors, age significantly predicted frequency of developmental speech errors, and severity of hypernasality significantly influenced frequency of compensatory articulation errors and passive cleft speech characteristics. Presence of cleft lip significantly impacted use of anterior oral cleft speech characteristics.

Future research should explore additional individual and treatment characteristics that may also influence speech production. Individual characteristics that should be examined include language functioning and its impact on use of developmental/phonological speech errors. Hearing status at time of speech evaluation and hearing history are other important variables related to speech functioning that should be examined, although it has historically been challenging to systematically document these characteristics. The impact of socioeconomic status on speech also warrants investigation. Several treatment characteristics should be investigated for their contributions to accurate speech articulation, including age at primary palate repair and primary palate repair type. Provision of speech therapy should be examined in terms of quantity and content of previous speech therapy services. Dental characteristics, as well as provision of Phase I orthodontic treatment, are relevant variables to study for children with cleft lip and palate. While these characteristics were not explored in the current study, they are acknowledged as possible contributors to speech production skills that should be examined in future projects.

Work on this study informed that three-year-old children with cleft palate had difficulty completing the sentence repetition task for the AESS. Many three-year-old children with speech recordings instead produced the sentences modified for younger children that consist of two-word and three-word phrases. The three-year-old sentences have been adopted for clinical use by several cleft teams, but little is known about how results from production of those sentences relates to other speech outcomes. However, it is the goal of cleft teams to conduct a full evaluation of articulation and resonance at the earliest age a child is compliant for standardized testing. Therefore, I would like for my next research study to examine speech accuracy and speech error usage during the sentence repetition task for the three-year-old sentences. I would like to conduct a longitudinal investigation of speech articulation for children with cleft palate who completed speech assessment using the three-year-old sentences at age three who later completed speech testing using the full AESS at ages four or five. I would like to explore how speech accuracy and rate of speech errors changes across these two time points (i.e., age 3 versus age 4-5), during an important period of speech development in children with cleft palate.

Summary and Conclusion

Children with CP+/-L presented with speech accuracy that differed across manners of production. As expected, nasal phonemes and glide phonemes were produced with the highest speech accuracy, followed by stop consonants, which are the earliest developing class of high-pressure phonemes. Children demonstrated substantially reduced accuracy of production for fricative and affricate phonemes, as well as the later-developing class of liquid phonemes. Fricative phonemes demonstrated the strongest correlation with other high-pressure phoneme classes as well as production accuracy for liquids. Speech accuracy for fricative phonemes was influenced by severity of hypernasality, although age and status of secondary surgery did not influence speech accuracy for fricatives.

For place of articulation, children with CP+/-L demonstrated strongest accuracy of production for bilabial and velar phonemes, while phonemes produced with alveolar and palatal articulation demonstrated significantly reduced accuracy.

Children with CP+/-L used a variety of speech error types, with anterior oral cleft speech characteristics, compensatory errors, and developmental/phonological errors occurring most frequently across the sample. Several factors impacted the type of speech errors used, including cleft type, status of velopharyngeal function, and age. Cleft type influenced rate of anterior oral CSCs usage, severity of hypernasality influenced rate of compensatory articulation errors and passive CSCs, and age predicted use of developmental/phonological speech errors.

Speech accuracy during a sentence repetition task was strongly correlated with speech accuracy during a single-word naming task. This finding indicated these assessment tools demonstrate concurrent validity, therefore supporting use of the AESS to evaluate articulation skills in children with CP+/-L.

Speech accuracy and speech error usage are important outcomes to evaluate for children with CP+/-L. Further study of these articulation skills, as well as the individual and treatment characteristics that may influence these skills, is warranted.

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APPENDIX A

AMERICAN ENGLISH SENTENCE SAMPLE

	American English Speech Sample - Sentences				
Trost-Cardamone, J. (2012) Please do not share without permission		Target sound(s)	√ = correct; write error in position		
			I	M	F
1	**<u>M</u>**om n' A**<u>m</u>**y are ho**<u>m</u>**e	**/m/**			
2	Pu**<u>pp</u>**y will **<u>p</u>**ull a ro**<u>p</u>**e	**/p/**			
3	Buy **<u>b</u>**a**<u>b</u>**y a bi**<u>b</u>**	**/b/**			
4	A fly **<u>f</u>**ell off a lea**<u>f</u>**	**/f/**			
5	I lo**<u>v</u>**e e**<u>v</u>**ery **<u>v</u>**iew	**/v/**			
6	**<u>Th</u>**irty-two tee**<u>th</u>**	**/θ /**			
7	**<u>Th</u>**e o**<u>th</u>**er feather	**/ð/**			
8	A**<u>nn</u>**a **<u>kn</u>**ew no o**<u>n</u>**e	**/n/**			
9	Your **<u>t</u>**urtle ate a ha**<u>t</u>**	**/t/**			
10	**<u>D</u>**o it to**<u>d</u>**ay for da**<u>d</u>**	**/d/**			
11	**<u>L</u>**aura will ye**<u>ll</u>**	**/l/**			
12	Si**<u>ss</u>**y **<u>s</u>**aw Sally ra**<u>c</u>**e	**/s/**			
13	**<u>Z</u>**oey ha**<u>s</u>** ro**<u>s</u>**es	**/z/**			
14	**<u>Sh</u>**e wa**<u>sh</u>**ed a di**<u>sh</u>**	**/ʃ/**			
15	Wat**<u>ch</u>** a **<u>ch</u>**oo-**<u>ch</u>**oo	**/tʃ/**			
16	Geor**<u>g</u>**e saw **<u>G</u>**igi	**/dʒ/**			
17	We are ha**<u>ng</u>**i**<u>ng</u>** on	**/ŋ/**			
18	A **<u>c</u>**oo**<u>k</u>**ie or a ca**<u>k</u>**e	**/k/**			
19	**<u>G</u>**ive A**<u>gg</u>**ie a hu**<u>g</u>**	**/g/**			
20	Hurry a**<u>h</u>**ead **<u>H</u>**arry	**/h/**			
21	I **<u>sp</u>**y a **<u>st</u>**arry **<u>sk</u>**y	**/sp, st, sk/**			
22	**<u>R</u>**ay will a**<u>rr</u>**ive early	**/r/**			
23	We **<u>w</u>**ere a**<u>w</u>**ay	**/w/**			
24	We ran a long mile	For hyponasality			