

PROFILING SPEECH AND LANGUAGE OUTCOMES OF CHILDREN
WITH CLEFT PALATE AT 39 MONTHS OF AGE: EXAMINING
PREDICTORS AND IDENTIFYING SPEECH AND
LANGUAGE CHARACTERISTICS

by

Lauren Nicole Beckett

A thesis submitted to the faculty of
The University of Utah
in partial fulfillment of the requirements for the degree of

Master of Science

in

Speech-Language Pathology

Department of Communication Sciences and Disorders

The University of Utah

August 2017

Copyright © Lauren Nicole Beckett 2017

All Rights Reserved

The University of Utah Graduate School

STATEMENT OF THESIS APPROVAL

The thesis of Lauren Nicole Beckett

has been approved by the following supervisory committee members:

Kathy Chapman , Chair May 8th, 2017
Date Approved

Sean Redmond , Member May 8th, 2017
Date Approved

Stacy Manwaring , Member May 8th, 2017
Date Approved

and by Michael Blomgren , Chair/Dean of

the Department/College/School of Communication Sciences and Disorders

and by David B. Kieda, Dean of The Graduate School.

ABSTRACT

The purpose of this study was to examine the speech/language skills of children with cleft palate and their noncleft peers at 39 months, profile the speech/language outcomes of children with cleft palate at 39 months, and extend previous studies examining pre- and postsurgery speech/language skills that predict later speech/language outcomes of children with cleft palate at 39 months.

Participants included 66 children, 43 children with cleft palate and 23 noncleft children. Spontaneous speech/language samples were collected at 9 months, postsurgery (approximately 13 months), 21 months, and 39 months of age in the child's home during an interaction with the caregiver. Speech and language measures were calculated using computer software programs and hand calculations. Children were classified into one of the four speech/language outcome profiles using descriptive statistics.

Results of the between-group comparisons revealed the children with cleft palate had fewer consonant sounds, produced less accurate consonants for the majority of the place and manner categories, and had lower mean length of utterances than their noncleft peers. Within-group comparisons revealed the risk factors gender, maternal education, and resonance were associated with poorer speech outcomes for children with cleft palate at 39 months. The profile normal velopharyngeal mechanism and delayed speech and/or language had the highest membership (41%). Correlations between pre- and postsurgery measures and later speech/language outcomes at 39 months revealed negative

correlations between 9 month predictors and all outcome measures. All other predictors were positively correlated with the speech outcome measures at 39 months. True consonant inventory and stop production measures at 21 months were the best predictors of the profile normal velopharyngeal mechanism and normal speech/language.

These results suggest that children with cleft palate have poorer speech/language outcomes than noncleft peers at 39 months of age. There is a need for children with cleft palate to receive earlier speech/language intervention to help them catch up with their noncleft peers. Finally, the strongest correlations were found between true consonant inventory and stop production at age 21 months, suggesting that 21 months is the best predictive age for speech and language outcomes at 39 months.

TABLE OF CONTENTS

ABSTRACT	iii
LIST OF TABLES	vii
LIST OF FIGURES	viii
ACKNOWLEDGEMENTS	ix
INTRODUCTION	1
REVIEW OF LITERATURE	5
Prelinguistic Development of Children With Cleft Palate: Presurgery	5
Speech Development of Children With Cleft Palate: Postsurgery	13
Relationship Between Prelinguistic Development and Later Speech Outcomes in Children With Cleft Palate.....	22
Language Skills in Children With Cleft Palate.....	26
Variable Outcomes.....	29
STATEMENT OF THE PROBLEM	36
METHOD	39
Participants.....	39
Data Collection Method.....	40
Data Analysis	43
Statistical Analysis.....	45
RESULTS	49
Question One	49
Question Two.....	50
Question Three.....	51
Question Four.....	52
Question Five.....	53
DISCUSSION	64

Speech Skills of Children With Cleft Palate Compared to Noncleft Peers	64
Predictors of Speech Outcomes in Children With Cleft Palate at 39 Months of Age	68
Speech Outcome Profiles.....	71
Variables that Predict Outcomes.....	74
Limitations of the Study.....	77
Clinical Implications.....	79
Conclusions.....	80
APPENDIX.....	82
REFERENCES	84

LIST OF TABLES

Tables

1. Participant Characteristics for Children with Cleft Palate and Noncleft Children	48
2. Mean (<i>M</i>), Standard Deviation (<i>SD</i>), Median (Med), Range, Significance Levels (<i>p</i>), and Effect Size (<i>d</i>) for Speech Outcome Measures of Children with Cleft Palate and Noncleft Peers at 39 Months	55
3. Mean (<i>M</i>), Standard Deviation (<i>SD</i>), Median (Med), Range, Significance Levels (<i>p</i>), and Effect Size (<i>d</i>) for Place of Production Measures of Children with Cleft Palate and Noncleft Peers at 39 Months	56
4. Mean (<i>M</i>), Standard Deviation (<i>SD</i>), Median (Med), and Significance Levels (<i>p</i>) for Manner of Production Measures of Children with Cleft Palate and Noncleft Peers at 39 Months	57
5. Mean (<i>M</i>), Standard Deviation (<i>SD</i>), Median (Med), Range, Significance Levels (<i>p</i>), and Effect Size (<i>d</i>) for Language Outcome Measures of Children with Cleft Palate and Noncleft Peers at 39 Months	58
6. Percentage Totals (%), Number of Participants (<i>N</i>), Significance Levels (<i>p</i>), and Test Statistics (χ^2) for Speech Status Based on Risk Factors of Children with Cleft Palate at 39 Months	59
7. Estimate of Coefficients and Standard Errors for Speech and Language Outcome Measures Based on Early Predictors for Children with Cleft Palate.....	62
8. Odds Ratio Point Estimations and 95% Confidence Intervals (<i>CI</i>), and Coefficient Estimation and Standard Error (<i>SE</i>) for Speech Outcome Profiles Based on Early Predictors in Children with Cleft Palate at 39 Months.....	63

LIST OF FIGURES

Figures

1. Percentage of children with cleft palate in each of the four speech/language outcome profiles60
2. Percentage of children with delayed speech, delayed language, or delayed speech and language for Profile 2, normal VP mechanism + delayed speech and/or language60
3. Percentage of children with delayed speech or delayed speech and language for Profile 3, questionable VP mechanism + delayed speech and/or language61

ACKNOWLEDGEMENTS

I would first like to express my sincerest thanks to my thesis chair and mentor, Dr. Kathy Chapman. I cannot thank you enough for the countless hours and late nights spent reading articles, analyzing data, writing and rewriting this thesis, and for sharing your passion for research with me. You have pushed me to be a stronger writer and a more analytical thinker. I could not have picked a more compassionate, knowledgeable, and supportive mentor than you. I would also like to acknowledge the additional members of my committee, Drs. Stacy Manwaring and Sean Redmond for your guidance and support throughout this process. Thank you for sharing your research expertise and pushing me out of my comfort zone. Your feedback and comments have been invaluable and this thesis would not be what it is today without each of your thoughtful critiques. I would like to thank Dr. Mary Hardin-Jones for answering all of my emails, and supporting me throughout this process.

Additionally, I thank Drs. Kathy Chapman and Mary Hardin-Jones for phonetically transcribing all of the speech and language samples, and Kelli Halter and Julie Shultz for collecting the speech and language samples. I would like to thank all of my fellow research assistants, and the volunteer students who helped me with data analysis. I especially appreciate all of the children and families who graciously participated in this study.

Finally, to my friends, Emma Logdahl, Lindsay Helderman, and Meg Rosales,

and boyfriend, Donald McPartland, who helped me with the editing process and whose constant words of encouragement kept me going. To my parents, Sue and Dirk Beckett, for provided me with unwavering support and continuous encouragement throughout my years of study and the process of researching and writing this thesis. Thank you for always telling me I could do anything; this accomplishment would not have been possible without you.

INTRODUCTION

Clefting impacts children's speech from an early age. Prior to palate repair, the structural deviations resulting from the cleft cause changes in speech acquisition due to oral-nasal coupling and the inability to impound air pressure for oral consonants. Without surgical intervention and speech-language intervention, these deficits often lead to delays in language and speech development. Notably, delays in lexical development and changes in place or manner of phoneme production, especially for obstruents, are most frequently seen. Hutters and Brøndsted (1987) found the changed place of articulation to be posterior to the site of velopharyngeal (VP) closure (e.g., glottal stop for /p/) or posteriorly placed within the oral cavity (e.g., velar fricative for /s/) (Chapman & Willadsen, 2011). Trost (1981) suggested these backed patterns of articulation be referred to as "compensatory articulations" (CAs) because the child with cleft palate¹ was attempting to compensate for a structural deficit. Grunwell and Russell (1987) initially suggested that while the cause of speech delays in children with cleft palate is the actual cleft, the structural impact on phonological learning may be more influential in speech acquisition. Difficulties in phonological learning can manifest in smaller consonant inventories, sound preferences, and collapsing of sound categories, all of which are referred to as "cleft-type phonological processes" (Harding & Grunwell, 1998).

¹ The term *cleft palate* is used to include children with a cleft palate with or without a cleft lip. A cleft lip does not impact speech in the same way as a cleft palate. We are only interested in the impact of the repaired palatal cleft on speech production and thus the term cleft palate will be used throughout this paper.

Over the past 50 years, numerous group studies have examined the deficiencies in the speech and language skills of children with cleft palate (see Chapman et al., 2008 for a review). The limitations of these works include a lack of noncleft comparison group, limited longitudinal data, and little information about speech and language outcomes. For example, some speech problems in children with cleft palate may be related to structural deficits post palatal repair. However, others may be phonological in nature, and others may be a combination of the two. While some researchers have suggested that the speech and language skills of children with cleft palate can be described according to four speech/language outcome profiles (listed below) based on their postsurgery speech and language performance, these profiles have not been validated with a large consecutive sample of children with cleft palate. The profiles include: 1) normal velopharyngeal (VP) mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, 4) questionable VP mechanism + normal speech and language (Scherer, Chapman, Hardin-Jones, & D'Antonio, 2005).

Additionally, studies have found evidence that important connections can be drawn between speech pre- and postsurgery performance (Chapman, 2004; Chapman, Hardin-Jones, & Halter, 2003; Jones, 2001). However, due to limited sample sizes and within group variability, more research is needed to define what early speech and language characteristics are related to the different profiles of performance.

The aims of this study were 1) to compare the speech of children with and without cleft palate at 39 months of age, 2) to examine risk factors (e.g., sex, episodes of otitis media (OM), resonance, therapy enrollment, and maternal education level) related to

delayed speech production at 39 months of age, 3) to determine how representative the different profiles of speech/language outcomes are of children with cleft palate at 39 months, 4) to examine early speech/language predictors of speech and language outcomes at 39 months, and 5) to examine early predictors of the four speech/language outcome profiles. To begin, a discussion of the literature is provided to describe what is already known about the presurgical speech, postsurgical speech, and pre- and postsurgical speech relationship. This is followed by a presentation of current research about the variable outcomes associated with postsurgical speech, and why a study examining all four speech/language outcome profiles would advance the literature on the outcomes of children with cleft palate.

Examination of the relationship between pre- and postsurgical speech production ability of children with cleft palate provides an opportunity to better understand how the developing speech system reorganizes itself after the mechanism is surgically repaired. It may also facilitate earlier identification of children who will later require speech and language intervention and/or additional surgical intervention. While we know that being born with a cleft palate impacts speech and/or language development in general, there is much to be learned about the specifics, such as how many children actually acquire normal speech and language without additional interventions, and what the risk factors and early predictors are related to speech and language outcomes. Categorizing these children into profiles based on their postsurgery speech outcomes would provide cleft palate and craniofacial management teams guidelines for treatment planning and “standards” against which they could compare other children. By addressing these study goals, we aimed to equip healthcare professionals with the tools necessary to identify

children with speech and language delays earlier, thus ensuring appropriate early intervention and management services could be provided prior to school entry.

REVIEW OF LITERATURE

Prelinguistic Development of Children With Cleft Palate: Presurgery

Babbling in babies is believed to be the first step toward word production. This vocal practice is important because the more frequently babies produce sounds and receive feedback, the more automatic their vocalization becomes, making it easier for them to execute each sound in a word (Stoel-Gammon, 1998). Feedback is a vital part of this sound development. Babies often repeat the same sound, which helps them learn the articulatory movements necessary to produce that sound. As a result, a baby's first words will typically include sounds that have been frequently babbled (Stoel-Gammon, 1998; Vihman, Ferguson, & Elbert, 1986; Vihman, Macken, Miller, Simmons, & Miller, 1985).

Babies born with cleft palate have the disadvantage of a constrained mechanism during the vocal practice stage. The open palate makes it difficult to vary production between oral and nasal sounds and can inhibit production of sounds requiring tongue-palate contact. These anatomical differences can cause the baby to selectively avoid difficult sounds, or to nasalize speech due to coupling of the oral and nasal cavities. In some cases, anatomical differences can even cause chronic middle ear disease, which can independently lead to an impairment in the baby's ability to hear both his or her own speech as well as others'. These factors can affect the baby either in isolation or in combination. Ultimately these differences impact which sounds are produced and how they are integrated into the developing lexicon (Peterson-Falzone, Trost-Cardamone,

Karnell, & Hardin-Jones, 2006)

Early on, researchers did not think there should be a focus on presurgery speech in children with cleft palate. They believed speech at that age was not meaningful. However, over the past 20 years, more studies focusing on children prior to surgical repair have led to a greater understanding of the impact clefting has on early vocal development (Chapman et al., 2001; Grunwell & Russell, 1987; Hardin-Jones & Karnell, 2010; Peterson-Falzone) were some of the first researchers to include presurgical measurements. Although their focus was to determine how long after palatoplasty vocalizations were influenced, their first measurement occurred prior to surgery, suggesting the children's presurgical speech was an important factor in their postsurgical development.

Chapman (1991) examined the vocalizations of five children with cleft palate prior to surgery compared to five noncleft children matched for age and sex. Spontaneous speech and language samples were collected in the children's homes while interacting with their mothers. The children's ages ranged from 12 months to 14 months at the time of testing. The children with clefts produced sounds primarily with a glottal placement (42% of productions), whereas the noncleft children produced primarily alveolar/palatal sounds (68% of productions). With regard to manner, the children with clefts produced primarily fricatives, specifically /h/ (38% of productions), compared to the noncleft children who produced more stops (71% of productions) than any other class of sounds. Both groups had a similar number of sounds in their consonant inventories. However, the noncleft children produced almost twice as many consonants during the spontaneous speech and language sample recording.

Comparisons were also made between the two groups for frequency and type of syllable productions. The children with cleft palate produced fewer multisyllabic productions (median = 16%) than their noncleft peers (median = 33%). In addition, their number of reduplicated (e.g. /bababa/) and variegated (e.g. /patiku/) multisyllabic productions was slightly lower than that of their noncleft peers (about 10% lower for each category). Overall, the children with cleft palate exhibited a preference for nasals, glides, and the glottal fricative [h] in their phonetic inventories.

In 1994, Lohmander-Agerskov, Soderpalm, Friede, Persson, and Lilja studied 35 children with cleft palate (with or without cleft lip), whose soft palates were repaired at the time of examination, but hard palates were unrepaired. The researchers analyzed the place and manner of pre-speech productions. Contrary to the findings of Grunwell and Russel (1987) and Chapman (1991), they found the majority of the children with clefts had supraglottal articulations. The only factor that was significantly correlated with place of articulation was the type of cleft a child had. Children with cleft palate and an unrepaired hard palate used posteriorly placed articulation more frequently, whereas children with only a soft palate cleft or no cleft used anteriorly placed sounds most frequently. Lohmander-Agerskov et al., (1994) also found a nonsignificant association between the size of the hard palate cleft and the frequency of anterior sounds in which children with smaller clefts had more anterior sounds. This finding supported Chapman's (1993) previous work suggesting that children with cleft palate more frequently substituted a sound with a more backed place of production in comparison to noncleft children.

Chapman, Hardin-Jones, Schuelte, and Halter (2001) expanded on the work of

Lohmander-Agerskov et al., (1994) by comparing the prelinguistic vocal development of 9-month-old babies with unrepaired cleft palate and their age-matched noncleft peers. This study also extended the results of Chapman (1991) by increasing the sample size and the range of speech measures examined. As noted in the earlier study, the babies with clefts produced significantly fewer stops, oral stops, and glides than the noncleft babies. Statistically significant differences were found in the number of velar and glottal productions for babies with cleft palate. The noncleft children did produce a slightly higher number of vocalizations during the sampling period; however, those differences were not statistically significant.

In contrast to the study conducted by Chapman (1991) which analyzed reduplicated versus variegated babbling, Chapman, Hardin-Jones, Schulte, and Halter's study (2001) included a different analysis of babbling development in addition to a phonetic analysis. Babbling measures included canonical babbling ratio (CBR), (i.e., number of canonical syllables divided by the total number of syllables (Oller, Levine, Cobo-Lewis, Eilers, & Pearson (1998)), true canonical babbling ratio (TCBR), (i.e., number of true canonical syllables divided by the total number of syllables) using the Stoel-Gammon (1989) definition of true consonants in which glides and glottals are excluded in the true consonant count (Chapman et al., 2001), percentage of babies who had reached the canonical babbling stage, and utterance types/syllable sequences. Children with clefts had lower CBRs as well as lower TCBRs. The number of babies with cleft palate who reached the canonical babbling stage was only about one-half (57%), compared to almost all (93%) of the noncleft babies.

The findings of Chapman, Hardin-Jones, Schulte, and Halter's (2001) study did

not show any significant differences for utterance type/syllable sequences between the cleft babies and the noncleft babies. Although these findings did not reach significant levels, the babies with cleft palate did produce a greater number of CV syllables, whereas the noncleft babies produced a greater number of CVCV syllables. Chapman et al. (2001) explained that although these findings appear contradictory, the CBR does not account for length of the babbled utterances. Also, in order to count as a canonical syllable for CBR, the utterance must include a supraglottal consonant, whereas the syllable length count (number of CV and CVCV syllables) did not exclude glottal consonants. Overall, the most noteworthy finding of this study was the significant discrepancy between the number of noncleft babies who had reached the canonical babbling stage (almost all babies) compared to only half of the cleft palate babies who had reached the stage. These results highlighted the vital role that structural differences, such as palatal clefting play in the development of speech beginning as early as 9 months of age.

Willadsen and Albrechtsen (2006) described the babbling of Danish toddlers with and without cleft palate. They found a difference between the groups for place and manner of articulation, suggesting a structural influence. The children with cleft palate produced significantly fewer oral stops and more nasals than their noncleft peers. The studies by Chapman (1991), Chapman et al. (2001), and Hutters et al. (2001) reported even lower percentages of oral stops than Willadesen and Albrechtsen's, but this difference can likely be explained by methodological differences and variable timing of surgery protocols across studies. As in earlier studies, the children with cleft palate produced fewer alveolar stops (Chapman, 1991; Chapman et al., 2001; Hutters et al., 2001; Lohmander, Olsson, & Flynn, 2011). However, unlike in earlier studies, these

children also produced more velar consonants than their noncleft counterparts (Chapman et al., 2001; Hutters et al., 2001). The authors suggested this finding was likely related to the babies with cleft palate showing a preference for velar sounds over alveolar sounds as they had undergone closure of the soft palate cleft at an earlier age (and the hard palate was unrepaired). As a result, the children were able to create closure behind the residual cleft resulting in the velar placement. This phenomenon of producing more velar than alveolar consonants has been documented before in children with residual clefting of the anterior hard palate (Lohmander-Agerskov et al., 1995, 1996, 2002).

Labial consonants were produced at a similar rate in the children with cleft palate and the noncleft children (Willadsen & Albrechtsen, 2006). A similar number of children from the cleft and noncleft groups had reached the canonical babbling stage, in contrast to the findings of Chapman et al. (2001) where only slightly over one-half of the children with cleft palate had reached the canonical babbling stage. Willadsen and Albrechtsen (2006) attributed these discrepancies to the age difference in the two groups and possibly to the fact that the Danish babies had undergone early soft palate repair (at 4 months of age). The children in their study were 11 months old rather than 9 months old as in the Chapman et al. (2001) study. Eleven months is on the older side of the age range for typical acquisition of canonical babbling and may have contributed to their better outcomes.

In a study by Chapman et al. (2008), the impact of both age and lexical status on the speech outcomes of preschool aged children with cleft palates was examined. The participants were 40 children between the ages of 33 months to 42 months. The children were split into two groups based upon vocabulary size at the time of palatoplasty. Group

1 included 20 children who were less lexically advanced (i.e., had fewer than five words as reported by their parents using the MacArthur Communicative Development Inventories [CDI; Fenson et al., 1993]) and also happened to have earlier surgery by an average of 1 month. Group 2 also included 20 children; however, these children were more lexically advanced (i.e., produced five or more words as reported by the parents using the CDI) and had later palatal surgery. Spontaneous speech and language samples were recorded and compared using the following speech production measures: size of consonant inventory, total consonants correct, percentage correct for manner of articulation categories, compensatory articulation usage, number of stable consonants, and percentage occurrence for CAs. Next, each sample was rated for articulatory proficiency and resonance using direct magnitude estimation (DME) ratings.

The results indicated a difference on four of the speech production measures. Children from Group 1 (i.e., those who produced less than five words and had early palate repair) exhibited larger consonant inventories with more accurate production of nasals and liquids. According to the DME ratings, listeners assigned higher articulatory proficiency scores and lower resonance scores (i.e., less hypernasality) to this group of children. Further examination revealed children from Group 1 were also rated as having more “normal” speech development. Overall, these findings suggest that children who are less lexically advanced and younger at the time of primary palatal repair have better speech, including articulation and resonance, at 3 years of age than their peers who are more lexically advanced and older at the time of surgery. While this study provided data on speech differences related to the timing of palatal surgery, the study did not include a noncleft comparison group.

The literature on speech development of typically developing children during the 1980s and 1990s “reported a correlation between frequency of occurrence of babbling, (consonant) diversity in babbling in the later babbling period, and the acquisition of early speech” (Willadsen & Albrechtsen, 2006, p. 190). In addition, acquisition of canonical babbling between 6 and 10 months of age has proven to be a strong developmental milestone for typically developing children. While it is unknown exactly how many children with cleft palate reach the canonical babbling stage on schedule, based on the work of Willadsen and Albrechtsen (2006), it seems that by 11 months of age, babies with early soft palate closure have similar canonical babbling frequency to noncleft babies. On the other hand, there is significant evidence to suggest that children with cleft palate differ from their typically developing peers in both place and manner of articulation of consonants. It can be inferred that these differences are a direct result of the structural deviation present during the early prelinguistic developmental stage for the children with cleft palate.

In order to determine if any presurgical speech characteristics were predictive of postsurgical speech outcomes, a good understanding of the speech characteristics of children with cleft palate prior to palatal surgery was necessary. The research presented above provided this. However, the studies primarily described the speech differences that occurred, and in some cases, provided a probable cause for the deviation from typical speech development. These studies did not address how early differences might affect the child’s speech acquisition later in life. When analyzed in conjunction with the postsurgical speech productions of children with cleft palate, these findings took on a new importance in identifying children who may later require further speech intervention

and/or additional surgery.

Speech Development of Children With Cleft Palate: Postsurgery

Some of the first studies examining the speech development of children with cleft palate focused on postsurgery differences between cleft and noncleft children. Bzoch (1965) and Philips and Harrison (1969) both completed cross-sectional studies of preschool aged children examining the differences between the speech of children with cleft palate and their noncleft peers. Each study employed different methods to evaluate speech, and both studies found the children with cleft palate were delayed relative to their noncleft peers. These researchers found that children with cleft palate were not as accurate in their speech sound production at age 5 as the noncleft children were at age 3. The types of speech errors produced by children with clefts were primarily omissions (four times more frequent than noncleft peers), substitutions, and distortions (both two times more frequent than noncleft peers). The children with cleft palate showed more errors on all sounds, particularly on stops, fricatives, and clusters, and frequently substituted glottal or pharyngeal sounds for stops, fricatives, and affricates (Bzoch, 1965). Philips and Harrison (1969) recommended that all children with cleft palate, specifically those with clefts of the soft palate, receive speech therapy prior to preschool entry to encourage them to reach their full potential with regard to speech development.

Many studies have used longitudinal data focused on children from as young as 2 years to as old as 18 years of age to explore the speech development differences in children with cleft palate (Grunwell & Russell, 1987; Karnell & Van Demark, 1986; Riski, 1979; Van Demark, Morris, & VandeHaar, 1979). Each of these studies addressed different areas of speech development, but reached similar conclusions. All babies

showed increases post palatoplasty in size of consonant inventory and use of multisyllabic utterances within the first 2 months postoperatively (Grunwell & Russell, 1987). In addition, children with cleft palate produced a greater number of errors than noncleft peers, and the children were more likely to have errors which persisted through adolescence (Grunwell & Russell, 1987; Karnell & Van Demark, 1986). Residual errors in production were seen as late as 18 years of age and affected stops, fricatives, and affricates (Van Demark, Morris, & VandeHaar, 1979). These errors were likely related to a compromised VP mechanism (Van Demark et al. 1979). These studies provided a substantial knowledge base regarding speech development of children with cleft palate. However, they primarily focused on describing errors in terms of manner categories. In addition, they are outdated and may not be representative of the more recent improvements in cleft palate management (i.e., earlier surgery, improved surgical techniques, aggressive otological management, etc.). For example, in the older studies, the typical timing of palatal surgery was around 18 to 24 months (Peterson-Falzone et al., 2008), whereas based on the survey conducted by Katzel, Basile, Koltz, Marcus, and Giroto (2009), the majority of surgeons (74%) registered with the American Cleft Palate-Craniofacial Association today reported they perform one stage palatal repairs when the child is between age 6 and 12 months.

Estrem and Broen (1989) were some of the few investigators to examine the phonology of early word productions of babies with cleft palate. Their analysis examined the phonetic characteristics of the first 50 words acquired by five children with cleft palate (four with repaired cleft palate), to identify the impact of clefting on lexical selectivity. In comparison to noncleft peers, children with cleft palate produced more

words beginning with sonorants (i.e., nasals, glides, liquids, and vowels) and fewer beginning with obstruents (i.e., fricatives, affricates, and oral stops). In contrast to many other studies, which suggested backing was a common phonological process employed by children with cleft palate (Chapman, 1993; Chapman & Hardin, 1992; Estrem & Broem, 1989; Willadsen & Albrechtsen, 2006), Estrem and Broem suggested children with cleft palate were using the extremes of the vocal tract. They found that children with cleft palate were not beginning words with alveolar, palatal, and velar sounds. Rather, they were producing more labial and glottal sounds as opposed to the noncleft children who had more coronals in the word-initial position. The authors noted, “the difference was most striking at earlier word levels but continued throughout the 50-word level (p. 5).” When accuracy of word- initial sounds was compared, the findings indicated a higher percent accuracy by the noncleft children compared to the cleft children (71.2% versus 53.6%). The authors suggested the discrepancy in production ability between cleft and noncleft children might have been related to early word choice, which would have been less linguistically diverse in the cleft children, or to the level of parental responsiveness, which could have been decreased due to poor intelligibility.

In another study of lexical selectivity, Willadsen (2013) compared the early lexicon of 34 Danish children with cleft palate (17 of whom still had an open residual cleft of the hard palate) and 35 age-matched peers at 18 months of age. Consistent with the findings of Estrem and Broen (1989), Willadsen found that children with cleft palate produced more word-initial nasals and glottals, and fewer word-initial oral stops and alveolars. In addition, the results showed that children with clefts were less accurate during production of initial consonants. In contrast to Estrem and Broen (1989),

Willadsen (2013) reported no selection bias toward sonorants since there was only a difference in the number of nasals and not in the number of glides, liquids, or vowels. Willadsen did not find that Danish-speaking children with clefts produced more word-initial labials and glottals (Estrem & Broen, 1989) but acknowledged the possibility of a language-specific or methodological difference to account for the disagreement between the two studies.

To provide further clarity of the early lexical characteristics of toddlers with cleft palate, Hardin-Jones, and Chapman (2014) examined 37 toddlers with cleft palate and 22 noncleft toddlers to determine whether they differed in expressive vocabulary size and lexical selectivity. The vocabulary size of toddlers at 13 months was similar for both groups. A trend toward smaller expressive vocabularies began to emerge by 17 months but was only found to be significant between 21 and 27 months of age. Findings for word initial lexical selectivity revealed that 70% of the cleft group demonstrated a preference for words beginning with sonorants compared to only 32% of the noncleft group. In addition, the cleft group accurately produced significantly fewer word-initial consonants with the majority of the errors occurring on obstruents. Hardin-Jones and Chapman explained that toddlers with cleft palate began to fall behind their noncleft peers for size of expressive lexicon by 17 months of age. They continued to lag behind for each of the subsequent ages even though the differences were only significant between 21 and 27 months. They also suggested that the types of sounds present in the child's inventory may impact lexical development. As for lexical selectivity, the findings from this study support those previously identified in Estrem and Broen (1989) and Willadsen and Albrechtsen (2006) that although both groups of toddlers had a similar number of sounds

in their inventory, the American English-speaking and Danish-speaking toddlers with cleft palate produced more sounds classified as sonorants, and the noncleft toddlers produced more sounds classified as obstruents.

In addition to lexical characteristics, children with cleft palate differ from their noncleft peers in other areas of speech production. Salas-Provance, Kuehn, and Marsh (2003) examined four babies with repaired clefts and four noncleft age-matched peers at 15 months of age. They found the babies with cleft palate had a more limited phonetic repertoire (about half as many phonemes) and more limited phonetic variations in their syllables than the noncleft babies, which was most evident in CV syllables. Syllable structure use was equal for both groups with CV syllables occurring most frequently, followed by isolated vowels. This study found all of the babies, cleft or not, were already beyond the canonical babbling stage, indicating CBR would be most informative earlier than 10 months of age.

Jones, Chapman, and Hardin-Jones (2003) examined speech production before and after palatal surgery. Their study included 14 children with cleft palate and 14 noncleft children, and they addressed the speech changes that occurred presurgery to postsurgery and how the children with cleft palate compared to their noncleft peers at 17 months and approximately 5 months postsurgery. When analyzing the pre- and postsurgery speech, they found the children with cleft palate produced more canonical syllables postsurgery compared to presurgery, although, interestingly, the increase was not noted for TCBR, suggesting the children with cleft palate were still producing glides in their canonical syllables. The consonant inventories of the children with cleft palate also increased in size from pre- to postsurgery. The discrepancy between TCBR and

consonant inventory size is likely explained by the idea that the children with cleft palate were adding new sounds to their inventory but still using them infrequently. As a result, a consonant would be counted as an “in consonant” for the consonant inventory measure. However, when calculating the TCBR, the total number of syllables containing that consonant may only be two, which would make the TCBR appear lower in comparison.

No statistically significant improvements were made pre- to postsurgery for place and manner, but oral stops increased 50% (particularly the bilabial stop [b]), and oral fricatives increased by a smaller margin. Glottal sounds decreased from the presurgery to postsurgery assessments at a level approaching significance.

When compared to noncleft peers, the children with cleft palate had TCBRs that were similar to same-age peers, indicating that the palatal repair was sufficient to allow the children with cleft palate to catch up on their production of canonical syllables. Additionally, by postsurgery (17 months), the children with cleft palate were found to have similar sized consonant inventories to their noncleft peers'. Finally, the noncleft children produced significantly more stops, oral stops, and alveolars, whereas the cleft children produced more nasals and glides. In summary, the children with cleft palate made gains in canonical syllables and consonant inventory but still showed deficits in production of alveolar place features and stops.

Both Jones, Chapman, and Hardin-Jones (2003) and Chapman, Hardin-Jones, and Halter (2003) found that children with cleft palate showed similar rates of true consonant acquisition at 17 and 21 months as compared to their noncleft peers. However, the accuracy of stop production by children at 21 months of age was lower than that of their noncleft peers. This lower production accuracy was seen for sounds with labial, dental,

alveolar, and velar place of production. This suggested that while children with cleft palate were making gains postsurgery, they continued to fall behind their noncleft peers for production of high pressure consonants (Chapman et al., 2003).

During the late 1980s to the early 1990s, there was a shift in the research, focusing on the phonological aspects of cleft palate speech. Phonological analyses were performed to distinguish any differences in the phonological development of children with cleft palate and their noncleft peers. Some of the investigators agreed that the error patterns of children with cleft palate could best be described using phonological rules (Berhardt, Doan, & Stoel-Gammon, 1995; Chapman, 1993; Chapman & Hardin, 1992; Hodson, Chin, Redmond, & Simpson, 1983; Lynch, Fox, & Brookshire, 1983; Powers, Dunn, & Erickson, 1990). However, others continued to argue that a phonological framework should not be used to describe the error patterns of children with cleft palate (Golding-Kushner, 2001; Peterson-Falzone et al., 1995; Trost, 1981).

Chapman and Hardin (1992) compared the phonetic and phonological skills of 10 two-year-olds with cleft palate, and 5 two-year-olds without cleft palate. No differences were found between children with cleft palate and their noncleft peers in the size of their consonant inventories. Significant differences were found for accuracy of production in nasals, liquids, and overall consonant production, with the noncleft children producing all sounds with higher accuracy than the cleft children. In addition, stops were also produced more accurately by the noncleft children; however, this difference was not found to be statistically significant. Next, they compared the use of phonological processes between the two groups and found significant differences in the use of nasal assimilation and backing for the children with cleft palate. However, only two children used one of the

processes at or above 20%, so their usage was not considered clinically significant. Finally, the frequency of CA's was examined. There was no significant difference between the two groups on this variable. Chapman and Hardin explained that backing is not unusual for children with clefts because of the nature of their structural deficit. While backing may be used to compensate for velopharyngeal dysfunction (VPD), at the time of assessment at age 2, it was unknown whether any of the children had VPD. As for nasal assimilation, the children with clefts may have struggled to produce more difficult oral sounds (i.e., stops and fricatives) and substituted a nasal sound at the same place of articulation.

Chapman (1993) further examined phonological process usage in older children with cleft palate to determine if there was a difference in the type or frequency of processes and how these children compared to noncleft peers. The findings suggested that phonological processes occurred at a higher frequency at 3 and 4 years of age for the children with cleft palate. However, by age 5, the children with cleft palate were more similar to their noncleft peers in the frequency of phonological processes produced. Analysis of the types of processes employed showed the children with cleft palate used the same set of processes as their noncleft peers with the exception of backing. Similar to the finding of Chapman and Hardin (1992), backing was more prevalent in the children with cleft palate. Again, backing was likely associated with an inadequate VP mechanism since the majority of the children using the process also expressed speech/resonance differences characteristic of VPD.

One study by Morris and Ozanne (2003) examined a group of 20 children with cleft palate to determine how their language, phonetic, and phonological skills differed at

3 years of age. The children were divided into two groups at age 2 years. One group of children had normal language development, while the other group of children had severely delayed expressive language development. The children were then assessed at 3 years of age, and comparisons were made on their comprehension, expressive language, and speech skills. Significant differences were found for total percent correct consonants and for plosives, nasals, fricatives, liquids, clusters, and cluster elements. In addition, the phonetic inventory of the delayed language group was significantly smaller. As for phonological characteristics, significant differences were found for seven phonological processes including nasal assimilation, final consonant deletion, cluster reduction, medial consonant deletion, glottal insertions, and nasal preference. It should also be noted that the normal language group also used phonological processes but only the children from the delayed group presented with the “atypical” processes (i.e., backing, glottal insertion, nasal preference, or initial/medial consonant deletion), which are most commonly associated with cleft palate. The authors concluded that the evidence from this study best supports the language/phonological disorder etiology in which impairments of linguistic organization lead to disordered communication skills.

These studies contributed substantially to the overall knowledge of speech outcomes for children with cleft palate, but they provided little insight into the factors that contributed to these speech differences. Based on these studies, it was unclear exactly how many children produced these errors in their postsurgery speech productions and how many “caught up” to their normally developing peers. Some recent studies have suggested the structural deficits of children with cleft palate could impact not only articulation but phonology as well. With concurrent data and stricter guidelines for

defining these speech delays/differences, children with cleft palate could have a more positive outlook and receive services that better meet their needs.

Relationship Between Prelinguistic Development and Later Speech

Outcomes in Children With Cleft Palate

Grunwell and Russell (1988) first hypothesized, “if these phonetic restrictions give rise to abnormal phonological patterns, it could be inferred that phonetic deviance has the potential to influence subsequent phonological development in cleft palate children” (p. 76). A comparison of speech pre- and postsurgery was necessary to understand how the speech of children with cleft palate changed once the mechanism was repaired. It was also important in distinguishing the presence or absence of a relationship between speech characteristics pre- and postsurgery. In typically developing children, researchers have found consistencies between syllable (Vihman, 1992) as well as consonant characteristics (Stoel-Gammon & Cooper, 1984) in prelinguistic verbalizations and later word productions (Oller, Weiman, Doyle, & Ross, 1976; Vihman, Ferguson, & Elbert, 1986; Vihman, Macken, Miller, Simmons, & Miller, 1985). Some of these same characteristics may likely be useful in identifying later speech and language delays in children with cleft palate as well. By defining these relationships, healthcare professionals could better identify at-risk children and begin managing, either surgically or behaviorally, their areas of deficit as early as possible.

Chapman, Hardin-Jones, and Halter (2003) examined the association between presurgery and postsurgery speech outcomes by focusing on the features present at specific time periods. They looked at presurgery (age 9 months) and immediate postsurgery (age 13 months) speech measures on the speech and language outcomes at

age 21 months in 15 children with cleft palate and 15 noncleft children. Children who produced more true stops at presurgery (9 months) scored significantly higher across all other speech production measures at 21 months (r values ranged from 0.59 to 0.72 suggesting a moderate to strong correlation). In addition, children who produced more true stops immediately postsurgery (13 months) had better speech and lexical development at 21 months including number of emerging consonants ($r = 0.70$), size of true consonant inventory ($r = 0.57$ for all utterances and $r = 0.62$ for lexical items only), and percentage of true stops ($r = 0.71$). Finally, size of true consonant inventory immediately postsurgery (13 months) was related to better speech and lexical measures at 21 months (r values range from 0.70 to 0.84 suggesting a strong correlation). Findings for noncleft children indicated that TCBR was the only variable related to later speech and lexical productions ($r = 0.64$) which supported the earlier research of Vihman et al. (1986) and Vihman and Greenlee (1987).

In a follow-up study, Chapman (2004) investigated if these relationships held true for older children with cleft palate. Unlike in the previous study, only children with cleft palate were included. Fifteen children were selected, all of whom were participating in a larger scale longitudinal study. Again, a comparison of the presurgery (age 9 months) and immediate postsurgery (age 13 months) speech measures were correlated with the speech and language outcome measures at a later age. Participants were compared at 39 months rather than 21 months. First, a moderate positive correlation was found between true stop production postsurgery (13 months) and mean length of utterance (MLU) at 39 months ($r = 0.52$). True stop production postsurgery was strongly correlated with number of different words (NDW) at 39 months ($r = 0.73$). Interestingly, TCBR and size of

consonant inventory presurgery (9 months) had a moderate to strong negative correlation with MLU at 39 months ($r = -0.62$ and $r = -0.71$, respectively), as did size of true consonant inventory presurgery and NDW at 39 months ($r = -0.52$).

Chapman provided two explanations for these findings. It may become apparent as the children age that VPD is present, or the children may not be receiving the early intervention services that are most beneficial. Chapman (2004) said “the impact of the early structural deficit is overshadowed by other variables in the postsurgery period that appear to have either a greater positive or negative effect on later communication skills” (p. 248). One problem with this study was that two of the children had extremely high TCBRs presurgery and some of the poorest speech and language outcomes at 39 months, making it appear that a negative correlation existed between TCBR presurgery and multiple speech and language outcome measures at 39 months. However, when the data from those two children were excluded, the only significant negative correlation remaining was between TCBR presurgery and MLU at 39 months. Therefore, additional research was needed to account for the small sample size and to further examine possible variables affecting later speech and language outcomes.

Scherer, Williams, and Proctor-Williams (2008) looked at a different set of pre- and postsurgery speech measures to determine if early detection of speech delays were possible for children with cleft palate and if these children differ at postsurgery from noncleft children. The study included a total of 26 children, half with cleft palate and half without. The data indicated that the children with cleft palate used less complex canonical babbling syllables (i.e., containing fewer stops) than the noncleft children at both 6 and 12 months of age, which supports the work of Chapman et al. (2001). The researchers

also noted a significant difference in vocabulary size and a nonsignificant difference in speech accuracy at 30 months. As for correlations between pre- and postsurgery speech measures, they found the children at 6 months who had a lower frequency of vocalizations made larger gains in consonant inventory size by the postsurgery 30-month measure ($r = -0.62$), meaning a moderate negative correlation was found. A similarly negative correlation was found by Chapman (2004) in which children with a lower TCBR at 9 months had a greater MLU at 39 months ($r = -0.62$). Scherer and colleagues (2008) also found a moderate negative correlation between the babbling frequency at 6 months and the vocabulary size at 30 months ($r = -0.58$). This contradicts the Chapman et al. (2003) finding that early canonical babbling was related to vocabulary size at 21 months.

Together, the studies that examined pre- and postsurgery speech outcomes and the relationships between them have found evidence of a link between early speech measures and later speech outcomes. This link was complex and involved a number of other variables outside of surgery alone. Researchers have identified some of the key markers which were related to speech delays in children with cleft palate. However, many of the studies were lacking in sample size and only compared results at two different timepoints. Chapman et al. (2003) and Chapman (2004) enhanced this knowledge base by looking at children across multiple timepoints and identifying a variety of speech and language measures. Further expansion of this work would provide more specific information about the age and speech measures most associated with later speech and language performance for children with cleft palate.

Language Skills in Children With Cleft Palate

Language performance in children with cleft palate received relatively little attention in comparison to speech acquisition. This lack of attention may have stemmed from early findings suggesting little difference between the language of children with cleft palate and their noncleft peers (Philips & Harrison, 1969b; Spriestersbach, Darley, & Morris, 1958). In addition, speech characteristics, especially those associated with VPD, were the most salient features noted by clinicians during treatment. More recently, research expanded to include a wider range of participants as well as a variety of methodologies to study language performance in children with children palate. A common conclusion was that “children with cleft palate are at risk for language delay,” (Chapman, 2008, p. 266). Therefore, measures of language performance and their influence on later speech and language development were considered.

Receptive language delays were reported by some investigators during the toddler and preschool years for children with cleft palate (Broen, Devers, Doyle, Prouty, & Moller, 1998; Chapman, Graham, Gooch, & Visconti, 1998; Fox, Lynch, & Brookshire, 1978; Jocelyn, Penko, & Rode, 1996; Morris, 1962; Nation, 1970; Philips & Harrison, 1969b). However, other researchers did not find delays, reporting that the children with cleft palate in their samples scored lower but not by a statistically significant margin (Long & Dalston, 1983; Neiman & Savage, 1997; Scherer & D’Antonio, 1995; Spriestersbach, Darley, & Morris, 1958). Chapman et al. (1998) reported that by school-age, children with cleft palate demonstrated age-appropriate comprehension. The two studies which indicated delays in the receptive language of children with cleft palate found hearing status during the first year of life to be one of the most influential factors

contributing to comprehension abilities (Broen et al. 1998; Jocelyn et al. 1996).

There was general consensus that young children with cleft palate had delayed word acquisition (Broen et al., 1998; Chapman et al., 2003; Hardin-Jones & Chapman, 2014; Scherer & D'Antonio, 1995). This delay was identified as early as 15 months of age (Broen et al., 1998), and it was unknown whether or not it continued into adolescence (Morris, 1962). Although children with cleft palate as a group were classified as having expressive language delays, individual children may be typically developing or even ahead of their same aged peers. As noted by Hardin-Jones and Chapman (2014), only 5 of the 40 children who participated in the Chapman et al. (2008) study examining lexical status based on timing of palatal surgery would have been characterized as delayed based on the criteria used to identify red flags of developmental language disorders by the FIRST WORDS Project ("Parent Report Measures," 2007). At the same time, Hardin-Jones and Chapman (2014) noted a tendency for the difference in lexical development to increase with increasing age, meaning the gap between the two groups grew over time. Chapman et al. (2001) identified slower onset of babbling, limited practice with supralaryngeal consonants, and auditory distortion impacting the feedback loop as possible causes of slow vocabulary development in young children with cleft palate. Broen et al. (1998) suggested VPD and hearing status as other possible contributing factors.

Examination of spontaneous language samples were used to compare the linguistic abilities of children with cleft palate in many studies. Two of the first studies, by Spriesterbach et al. (1958) and Morris (1962) found similar delays in mean length of response for children with cleft palate; however, structural complexity was not delayed

except for in the youngest children in one of the two studies. In the Morris (1962) study, the number of one-word responses, number of different words, and mean length of five longest responses were also significantly different compared to noncleft children. In the 1990s, Scherer and D'Antonio (1995) found differences in mean length of utterance (MLU) between the cleft and noncleft children, while many other studies reported children with cleft palate had MLUs similar to their noncleft peers (Broen et al., 1998; Jocelyn et al., 1996).

Pragmatics in children with cleft palate was considered a relative strength based on the work by Long and Dalston (1982). Warr-Leeper, Crone, Carruthers, and Leeper (1988) suggested preschoolers with cleft palate scored within normal limits on a standardized measure of pragmatics, the Test of Pragmatic Skills (Shulman, 1985). However, they found school-age children with cleft palate were delayed, but rather than attributing it to a pragmatics disorder, they hypothesized older children and adults with cleft palate simply produced shorter utterances. Chapman et al. (1998) used the model of conversational assertiveness created by Fey (1986) to examine pragmatics in preschool and school-aged children with cleft palate. No statistically significant group differences were found using the standardized test or conversational measures. But, when compared on an individual basis, 50% of the preschoolers and 20% of the school-aged children with cleft palate demonstrated a more passive style of conversation interaction, meaning, they were less likely to initiate a conversational turn. These findings were replicated in 2006 by Fredrickson, Chapman, and Hardin-Jones with a larger number of preschoolers with cleft palate and their noncleft peers (17 children in each group). They found 35% of preschoolers with cleft palate to be less assertive or less responsive than their noncleft

peers and also found the children with cleft palate produced fewer assertive utterances, were less likely to respond fully to caregivers, and produced more topic maintaining rather than topic extending utterances. Finally, the researchers found a positive correlation between level of conversational assertiveness and speech proficiency, indicating children with limited assertiveness had poorer articulation skills.

Children with cleft palate typically have had receptive language scores in the normal range, whereas expressive language scores were more variable. Many inconsistencies existed in the research due to a number of variables including the language skills examined, testing procedures, and the age at time of testing. The present study sought to expand on this by defining the prevalence of language disorders in a concurrent sample of children with cleft palate and examining the comorbidity of speech impairments or VPD with language delays.

Variable Outcomes

No current evidence is available about the prevalence of particular speech outcomes, including percentages and ranges of children with cleft palate who achieve normal speech, VPD, and speech and language delays. Also, very limited data exist describing the overall variability in speech and language of children with cleft palate. Bringing awareness to the number of children in a concurrent sample who fall into each speech outcome group could enhance management of these children's deficits while also ensuring all of their needs are simultaneously met. Previous studies have looked at different aspects of speech outcomes with varied results. Below were some areas of distinction.

Normal Speech

Many investigators predicted children with cleft palate had the capacity to reach normal speech after their structural abnormalities were surgically repaired. In 1973, Spriestersbach et al. estimated 50% of children with repaired cleft palates would spontaneously develop normal speech. Similarly, Bzoch (1997) suggested that the key component to normal speech and language development in the preschool years is having the best cleft palate team care prior to school entry. Blakeley and Brockman (1995) assessed children with cleft palates every 3 to 4 months until they reached age 5 and found that 88% of those children demonstrated normal resonance and articulation. However, this finding contradicted the majority of studies, which suggested a higher percentage of children were requiring speech therapy all the way into adolescence. Variability is the only shared finding present in these studies. Chapman (2008) outlined it best, stating that normal speech may develop immediately following surgical repair or after a period of speech therapy and orthodontic/surgical interventions for some children, and others may not ever exhibit totally normal, errorless speech.

Sell et al. (2001) in the United Kingdom described a clinical audit examining the speech outcomes of children born with unilateral cleft palate. They found over 80% of 5-year-olds had intelligibility or resonance issues, meaning less than 20% had normal speech. In the same study, they found only 47% of 12-year-olds had exhibited entirely normal speech. Although the percentage of children who had been enrolled in speech therapy was reported in the study, it was not clear how many of the children with normal speech had previously undergone therapy. It was likely that many of these children had received therapy considering the majority of the children in each group had speech

therapy (68% of 5-year-olds and 60% of 12-year-olds). These numbers were similar to those reported in studies of children in the United States. Peterson-Falzone, Trost-Cardamone, Karnell, and Hardin-Jones (2006) made a similar assumption about younger children, stating “about 25% of cleft palate preschoolers who receive team care” should produce normal speech (p. 21).

None of the present studies specifically described the number of children with cleft palate who had normal speech prior to school entry. It was encouraging that some of these children were able to reach normal speech levels, likely with the assistance of therapy. However, the literature was lacking on the number of children who developed normal speech following palatoplasty and the number of children who required minimal speech therapy to attain normal speech. In addition, the aim of earlier palatal surgery was primarily to provide a normal mechanism for infants to begin to develop speech, but little basis existed on which to substantiate the claims that surgery alone was enough to produce normal speech.

Velopharyngeal Function

Velopharyngeal dysfunction (VPD) in children with cleft palate is typically associated with a number of speech characteristics, including abnormal resonance resulting in hypernasality, nasal air emission, and nasal or facial grimace (Sell, 1996). Reports on the number of children achieving adequate VP function following primary surgery have varied. Morris (1973) and Spriestersbach et al. (1973) both proposed about 25% of children had VPD following surgery. Enderby and Emerson (1995), however, stated VPD varied from 5% to 40% of children with cleft palate. Much of this variability could be accounted for by differences in methodology and assessment protocols,

especially in the criteria for determining what constitutes normal speech. Peterson-Falzone and Graham (1990) presented a good example of this difference when they reported the prevalence of VPD to be 16% but 40% if more strict criteria were in place.

In the large-scale study mentioned above by Sell et al. (2001) in the United Kingdom, the number of 5- and 12-year-olds who had already undergone secondary VP surgery or were awaiting surgery were also reported. Initially, Sell et al. found only 16% of 5-year-olds and 20% of 12-year-olds had already received secondary surgery for VPD with only a few more children from each group still awaiting surgery. When accounting for the number of children with hypernasality rated in the moderate to severe range, they estimated 29% of 5-year-olds, and 32% of 12-year-olds had VPD due to inadequate primary surgical repair. The authors suggested that the fewer number of 5-year-olds (compared to 12-year-olds) requiring secondary surgery could be a result of improved surgical techniques and overall VP management.

In a more recent study, Hardin-Jones and Jones (2005) reported on the prevalence of hypernasality in their study of speech outcomes in preschool children residing in the United States of America. They found that 25% of the children had received secondary surgery to manage VPD, but when combined with the children who demonstrated moderate to severe hypernasality, that number grew to 37%. Hardin-Jones and Jones found a weak significant relationship between cleft type and the number of children with moderate to severe hypernasality (Cramer's V coefficient = 0.25), which indicated the children with more severe clefts were at more of a risk for VPD and/or speech delays. A weak significant relationship was also found between the age at primary palatal surgery and the number of children with moderate to severe hypernasality (Cramer's V

coefficient = 0.24).

As described above, research on the prevalence of VPD in children with cleft palate varied widely due to differing methodologies and criteria of diagnosis. It would be beneficial to add to this literature by defining the number of children with questionable VP function at age 39 months. Therefore, comparisons could be made between the number of children with questionable VP function and the number with other speech outcomes.

Speech Disorders

All children with cleft palate have differences in their development of speech, “including acquisition of the oral-nasal contrast, shift in ratio of glottal or supraglottal consonants, onset of canonical babbling, etc.” (Chapman & Willadsen, 2011, p.28). However, these changes do not impact each child’s speech and language development in the same manner. McWilliams et al. (1990) described the primary speech disorders associated with cleft palate, including abnormal 1) articulation (e.g., consonant production errors, nasal air emission, CAs); 2) resonance (e.g., hypernasality); and 3) speech quality (e.g., hoarseness). Not all children with cleft palate would exhibit one of these features, but the majority of children would have some type of difference or delay in speech development.

In the preschool years, children with cleft palate may have more speech sound errors than their peers, including omissions, substitutions, and distortions (Bzoch, 1965; Philips & Harrison, 1969a). These speech sound errors often negatively impacted the child’s intelligibility so listeners perceived them as poorer speakers; however, little research had shown how many children with cleft palate actually had these speech sound

or articulation based delays.

In the Sell et al. (2001) study previously described, speech outcomes for nasality, intelligibility, consonant errors, and residual need for speech therapy services were evaluated. The data showed high rates of unintelligibility at both ages (19% for 5-year-olds, 4% for 12-year-olds) when rated by unfamiliar listeners. They also found 34% of 5-year-olds and 17% of 12-year-olds had at least one serious consonant error with dentalization/interdentalization occurring most frequently for both age groups. Lastly, according to the data collected, 46% of 5-year-olds and 15% of 12-year-olds were judged as needing speech therapy but were not currently receiving any services. This finding was interesting because approximately two-thirds of the children from both age groups had already undergone speech therapy in the past.

A similar study in the United States by Hardin-Jones and Jones (2005) examined the prevalence of children with cleft palate who required speech therapy, had significant nasalization, and produced compensatory articulations. The 212 preschoolers examined ranged from 2 years 10 months to 5 years 6 months in age. They found approximately two-thirds of the children were currently or previously receiving speech therapy and at least one-quarter of the children were producing CAs. These findings were contrary to many previous studies, including Bzoch's (1997), where he assumed that proper management of cleft palate speech had the capability to correct communication disorders and provide future generations with better speech outcomes. The study by Hardin-Jones and Jones demonstrated the need for continued speech intervention to manage the severe speech sound delays of children with cleft palate, even with the frequent histories of speech therapy.

Co-occurring Disorders

Another underexamined speech outcome was the likelihood of a child with cleft palate having multiple co-occurring deficits. None of the current research on speech outcomes for children with cleft palate had specified the number of children who had both VPD and speech or language delays or VPD and no speech or language delays. The study by Hardin-Jones and Jones (2005) provided some evidence that overlap between the two profiles existed. Their findings approximated two-thirds of the children had received speech therapy at some point in time. Another 37% of the children had undergone secondary surgery to manage VPD, or were significantly hypernasal. Although it was not explicitly stated, it was likely some of the children with VPD had also required speech therapy to ensure correct placement for pressure consonants in order for an assessment of VP competency to even be made. Further evidence of comorbidity was provided by Pamploma, Ysunza, and Espinosa (1999), who studied children with cleft palate in Mexico and found that children with both CAs and VPD were more likely to also present with language delays compared to the children who had VPD with no CAs present.

STATEMENT OF THE PROBLEM

The preceding review of the literature provided preliminary evidence that the speech/language of children with cleft palate presurgery could provide insight into the later speech/language outcomes postpalatoplasty. Furthermore, there was evidence that children with cleft palate had variable speech outcomes and were at risk for speech/language delays. Some of these problems could be remediated with speech therapy or secondary surgery, and with others persisting into adolescence and adulthood. Clearly, in the case of children with cleft palate, many speech outcomes are possible following primary palatal surgery. Current studies on the outcomes of children with cleft palate focused on the average group performance for one to two specific speech or language measures. Additionally, these studies looked at children cross-sectionally and did not provide longitudinal data about how the same population of children developed from presurgery to postsurgery. Due to inconsistencies across the literature, a study outlining the consecutive breakdown of how many children with cleft palate have each type of speech outcome would provide useful information for professionals managing children with cleft palate.

This study compared the speech and language skills of children with cleft palate and their noncleft peers at 39 months. This age was chosen because by 39 months, typically developing children have acquired the majority of their consonant sounds, including most stop consonants (Smit, Hand, Freilinger, Bernthal, & Bird, 1990). This

production of stop consonants is important for diagnosis of VPD and perceptual problems associated with VPD (e.g., nasal emission on pressure consonants). Additionally, this study examined risk factors that may contribute to variability in speech/language skills at 39 months of age, including 1) sex, 2) maternal education level, 3) episodes of otitis media (OM), 4) resonance, and 5) therapy enrollment. Next, the number of children who fell into each of the four speech/language outcome profiles: 1) normal VP mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, 4) questionable VP mechanism + normal speech and language was calculated (Scherer et al., 2005). The four speech outcome profiles were further defined to determine what type of speech and language delays were most prevalent among children with cleft palate. Third, this study attempted to replicate and extend the findings of Chapman, Hardin-Jones, and Halter (2003) and Chapman (2004) with a few modifications to determine what age and speech measures were most predictive of later speech outcomes for children at 39 months of age.

Based on the review of the literature, it was predicted that children with cleft palate would perform worse than their noncleft peers for the majority of the speech and language measures. It was predicted that males and children with hypernasal resonance would have worse speech outcomes at 39 months. Since no other studies have categorized children with cleft palate based on their speech and language outcomes, it was difficult to predict which profile would have the highest membership. However, based on the high rates of hypernasality present in the Sell et al., (1999) study, it was predicted that the speech/language outcome profiles with questionable VP mechanism

would be the most common. Findings for the predictive questions were expected to extend the previous pre- and postsurgery relationship studies which have suggested that the number of stop consonants presurgery were the strongest predictors of later speech/language outcomes.

The following experimental questions were addressed:

- 1) How do the speech and language skills of children with cleft palate compare to those of their noncleft peers at 39 months?
- 2) Which risk factors (i.e., sex, maternal education, etc.) are most associated with disordered speech production skills for children with cleft palate at 39 months of age?
- 3) At age 39 months, what percentage of children with cleft palate fit into each of the four speech/language profiles?
- 4) What timepoint(s) and speech/language variables are most predictive of speech outcomes at 39 months?
- 5) What timepoint(s) and speech/language variables are most predictive of each of the four speech/language profiles?

METHOD

Participants

The participants included 66 children: 43 children born with cleft palate and 23 children without a cleft who participated in a multisite longitudinal study of speech and language development of young children with cleft palate from age 6 months to 39 months (Chapman, 2004; Chapman et al., 2008; Chapman, Hardin-Jones, & Halter, 2003; Chapman, Hardin-Jones, Schulte, & Halter, 2001). The inclusionary criteria for participants included complete cleft of the hard and soft palate (with or without cleft lip) and cognitive functioning within typical limits as measured by Bayley Scales of Infant Development (BSID), (Bayley, 1993). Exclusionary criteria included any known or suspected neurological impairments, an associated syndrome, or a sensorineural hearing loss.

Of the 92 children who participated in the longitudinal study, 66 were selected for this study. The children with cleft palate were selected based on the type of surgical repair they had (i.e., one-stage surgical repair rather than two-stage surgical repair) and their participation in the majority, if not all of the timepoint(s) examined in this study. An independent *t*-test showed no statistical differences between the cleft and noncleft children for exact age at 39-month session. The children were not similar for occurrence of OM due to the higher rates of OM in children with cleft palate (Paradise, Bluestone, & Felder, 1969; Stool & Randall, 1967; Rynnel-Dagoo, Lindberg, Bagger-Sjoback, &

Larson, 1992; Peterson-Falzone et al., 2010). Despite the differences in OM status, all children received aggressive otologic care (see Table 1 for additional information regarding participant characteristics).

Data Collection Method

The children with cleft palate participated in a presurgery session which occurred between ages 08;26 and 15;10 [months; days], and a postsurgery session which occurred between ages 9;22 and 19;7. The variation was related to differences in timing of surgery across children. All children were assessed between 2 weeks presurgery and no more than 6 weeks postsurgery. Additional sessions occurred at 9 months, 13 months, 17 months, 21 months, 27 months, 33 months, and 39 months for all participants. For this study, data from the 9 month/presurgery, postsurgery (mean age 14 months), 21 month, and 39 month sessions were analyzed.

All data collection sessions were conducted in the child's home with a parent and a trained research assistant present. Each session lasted approximately 60 to 90 minutes. A spontaneous speech/language sample was collected during a parent-child interaction at each timepoint. The samples lasted approximately 30-60 minutes or until 100 child utterances (9 months, 13 months, postsurgery, and 17 months) or 200 different words (21 months, 27 months, 33 months, and 39 months) were elicited. Tympanometry testing was also performed at each timepoint to assess middle ear function. Additional testing procedures were completed for each but for the purposes of this study, only the spontaneous speech/language samples and the PLS-3 (Zimmerman, Steiner, & Pond, 1992) at 39 months were used.

The spontaneous speech/language samples were collected with a Marantz portable

cassette recorder (PMD 430), Countryman wireless microphone (MEMWF05ETS), Telex receiver (RMR-70) and transmitter (WT60), and Panasonic video camera (Model AG188). The wireless microphone was clipped to the child's clothing approximately seven to ten inches from the child's mouth. It was clipped to ensure the child could move freely while playing and also provided a quality recording. The research assistant instructed each parent to interact naturally with his or her child in order to elicit the sample. Both the parent and child were recorded during the sample, and the interaction occurred while playing with a standard set of toys provided by the research assistant.

At the prelinguistic timepoints, only the mother's utterances were orthographically transcribed as the child's speech was nonmeaningful. As words began to emerge in the child's repertoire, both the mother and child's utterances were orthographically transcribed. Orthographic transcription was done on a computer using the Systematic Analysis of Language Transcripts (SALT V 6.1a) software (Miller & Chapman, 2000) then phonetically transcribed by Kathy Chapman or Mary Hardin-Jones, the primary investigators on the study. The samples were transcribed using the International Phonetic Alphabet (IPA) (1989) in addition to diacritic markings (Shriberg & Kent, 1982) and CA symbols as described by Trost (1981).

Metaphonological/infraphonological coding (Oller, 1986, 2000; Oller & Lynch, 1992) was used to capture the productions that could not be transcribed using IPA symbols made by the babies at early timepoints. Utterance boundaries were determined by the primary investigators using "breath groups," or pauses lasting 1 or more seconds (Lynch, Oller & Steffens, 1989; Oller & Lynch, 1992). Only utterances containing speech-like consonant or vowel segments were analyzed.

Interrater reliability was calculated for phonetic transcription agreement for 15% of the transcripts (randomly chosen across the timepoint(s) studied). Only sounds that were transcribed identically for place, manner, and voicing were considered agreements. Percent agreement scores were calculated by dividing the number of agreements by the total number of consonants produced in the sample (number of agreements and disagreements). The agreement scores for interrater reliability ranged from 84% to 91% and the mean score was 87%.

Transcribed samples were entered into the Logical International Phonetic Programs (LIPP) (Oller, 1990), which allowed for analysis of specific features of the transcribed data. These analyses are described further in the Appendix.

The language outcome measures were calculated from 150-utterance spontaneous speech/language samples using the SALT program (Miller & Chapman, 2000) at 39 months. This utterance length was chosen based on the research of Gavin and Giles (1996), which suggested samples containing 150 or more utterances led to reliability of greater than 75%. These measures included 1) the number of different words (NDW) produced in the spontaneous speech/language sample and 2) the mean length of utterance (MLU) in both words and morphemes, which was calculated by adding the number of words/morphemes in a 150-utterance sample and dividing by the total number of utterances. At 21 months, NDW was calculated from a 20-minute sample due to the large variation in total number of utterances. Gavin and Giles (1996) found over 70% reliability when calculating NDW from a 20-minute sample. The 20 minutes began 1 minute after the child's first verbalization. Interrater reliability was calculated for SALT coding agreement for 15% of the 39 month samples. The mean agreement score was

87%, and the range of agreement scores was 79% to 98%.

Resonance was determined based on consensus of the two primary investigators. Ratings were as follows: 1) normal, 2) mild hypernasality, 3) moderate hypernasality, or 4) severe hypernasality. Speech status (speech within normal limits or speech not within normal limits) was analyzed based on the normative data by Smit, Hand, Freilinger, Bernthal, and Bird (1990) in which children who produced more than two age-appropriate sounds with less than 75% accuracy were considered to have delayed speech.

Data Analysis

Experimental Question One

Between-group comparisons were made using independent sample *t* tests and Wilcoxon rank-sum tests to examine the speech and language skills of children with cleft palate compared to their noncleft peers at 39 months of age. Speech variables included 1) size of true consonant inventory (TCI), 2) stable consonants, 3) percentage of correct consonants, 4) percentage of consonant accuracy for all place and manner characteristics, 5) number of different words (NDW), and 6) mean length of utterance (MLU) for both words and morphemes. See the Appendix for a more descriptive explanation of each measure.

Experimental Question Two

Chi-square analyses were performed to determine which risk factors were most associated with disordered speech production skills for children with cleft palate at 39 months. The risk factors included 1) sex, 2) maternal education level, 3) episodes of OM, 4) resonance status, and 5) therapy enrollment. Speech production skills were categorized

as either within normal limits or not within normal limits based upon the speech status measure described above.

Experimental Question Three

Descriptive analyses were employed to show the percentage of children with cleft palate who fit into each of the four speech/language outcome profiles; 1) normal VP mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, and 4) questionable VP mechanism + normal speech and language (Scherer et al., 2005). Children rated as a three or four on the resonance scale (described above) were considered to have a questionable VP mechanism. Children rated as a one or two were considered to have a normal VP mechanism. Speech was categorized as within normal limits or not within normal limits based on the speech status measure described above. Language was assessed with PLS-3 (Zimmerman et al., 1992) scores at age 39 months, NDW produced, and MLU in morphemes computed through the SALT program (Miller & Chapman, 2000). The criterion of one standard deviation below the mean was used to identify delayed performance on the PLS-3 (Zimmerman et al., 1992) and MLU and NDW as defined by Miller (1981). Children were categorized as language delayed if they were delayed in two of three of the language measures.

Experimental Question Four

A linear regression analysis was performed to determine what early timepoint(s) and speech and language characteristics were most predictive of speech and language outcomes at 39 months. At the 9-month/presurgery and postsurgery timepoints, the

following measures were assessed: 1) true canonical babbling ratio (TCBR), 2) true consonant inventory (TCI), and 3) percentage of true stop consonants. At 21 months, the speech and language measures included 1) TCI, 2) stable consonants, 3) percentage of correct consonants, 4) percentage of true stops, 5) total correct stop production, and 6) NDW. The outcome measures at 39 months included 1) TCI, 2) stable consonants, 3) percentage of correct consonants, 4) percentage of true stops, 5) total correct stop production, 6) NDW, and 7) MLU in morphemes.

Experimental Question Five

A logistical regression analysis was performed to determine the timepoint(s) and speech and language variables which were most predictive of the four speech/language outcome profiles. The predictor variables were the same as listed above; however, the outcome measures were the four speech/language outcome profiles: 1) normal VP mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, 4) questionable VP mechanism + normal speech and language (Scherer et al., 2005). The same criteria described in question three were used to define the four speech/language outcome profiles.

Statistical Analysis

Independent sample *t*-tests (parametric) and Wilcoxon rank-sum tests (non-parametric) compared the differences in speech and language skills between the cleft children and noncleft peers. When variables were not normally distributed, the non-parametric test was used. This measurement determined if significant differences existed

between the speech and language skills of the two groups (Experimental Question One). To determine which risk factor variables were the best predictors of disordered speech production skills at 39 months, chi-square analyses were run for each individual dependent variable (Experimental Question Two).

Linear regression analyses using selected factors based on the Least Absolute Shrinkage and Selection Operator (LASSO) approach were used to identify the timepoint(s) and speech and language variables most associated with normal speech and language at 39 months (Experimental Question Four). The LASSO approach identified the subset of potential predictors that were truly informative for predicting the probability of the outcome variable at 39 months. This occurred by imposing some penalty in the regression model fitting which shrank the coefficients of those unimportant predictors to zero while retaining the important predictors. It should be noted that a predictor variable only had predictability on the outcome of interest if, and only if, its coefficient was nonzero. Thus, the final models included all important predictors with improved precision. Once the variables were selected, linear regressions were performed to determine the strength and direction of predictability for each predictor variable. Data were standardized to allow for comparisons between of each of the predictor variables.

To identify the timepoint(s) and speech and language variables most associated with the four speech/language outcome measures (Experimental Question Five), logistical regression analyses using selected factors based on the LASSO approach were used. The LASSO approach was again used to select the predictor variables of interest in a simultaneous way. The outcome measures (speech/language outcome profiles) were collapsed so each regression had a binomial outcome (i.e., predictor variables were used

to distinguish classification between two possible outcomes). The logistical regression was performed to determine the odds ratio (OR), which was the ratio between the probability that a child would fall into one profile versus the probability they would fall into a second profile. This distinguished which predictor variables could be used to predict if a child would fall into one of two speech/language outcome profiles.

Table 1
Participant Characteristics for Children with Cleft Palate
and Noncleft Children

	Cleft	Noncleft	Total
Sex			
Male	27 (63%)	14 (61%)	41
Female	16 (37%)	9 (39%)	25
Type of Cleft			
BLP	9 (21%)	0	
ULP	26 (60%)	0	
HSP	4 (9%)	0	
SPO	4 (9%)	0	
Maternal Education			
No High School	2 (5%)	1 (4%)	3
High School	15 (35%)	7 (30%)	22
Some College	10 (23%)	4 (17%)	14
Associate	1 (2%)	2 (9%)	3
Bachelor	14 (33%)	5 (22%)	19
Master	0 (0%)	4 (17%)	4
Doctorate	1 (2%)	0 (0)	1
Received Therapy			
Yes	26 (60%)	2 (9%)	28
No	17 (40%)	21 (91%)	38
Episodes of OM			
Median	4.5	2	
Range	0-Chronic	0-Chronic	
Age of Surgery (Mos.)			
Median	12		
Range	7-18		

Note. BLP = Bilateral cleft lip and palate; ULP = Unilateral cleft lip and palate; HSP = Cleft of the hard and soft palate; SPO = Cleft of the soft palate only; OM = Otitis media

RESULTS

Question One

The first statistical tests compared the speech skills of children with cleft palate to their noncleft peers at 39 months of age based on spontaneous speech and language sample findings. Results of independent t tests and Wilcoxon rank-sum tests revealed significant differences between the cleft and noncleft groups for true consonant inventory ($z = -3.478$; $p = <0.001$), number of stable consonants ($t = -3.674$; $df = 64$; $p = <0.001$; $d = -1.014$), and percentage of correct consonants ($z = -3.936$; $p = <0.001$). Analysis of effect size with Cohen's d for the normally distributed variable revealed a large effect for the number of stable consonants ($d = -1.014$). In all cases, the noncleft children performed better than the children with cleft palate. The children with cleft palate had an average of two less consonants in their inventory and an average of three fewer stable consonants than the noncleft children (Table 2).

Comparisons between the cleft palate and noncleft groups using independent t tests and Wilcoxon rank-sum tests revealed significant differences for all place of production features at 39 months. The children with cleft palate produced significantly fewer labial ($z = -3.264$; $p = 0.001$), dental ($z = -3.120$; $p = 0.002$), alveolar ($t = 3.815$; $df = 64$; $p = <0.001$), palatal ($t = -4.562$; $df = 64$; $p = <0.001$), and velar ($z = -2.967$; $p = 0.003$) sounds than their noncleft peers. Effect sizes were calculated for each of the normally distributed measures with Cohen's d . Results indicated a large effect for both

alveolar ($d = -1.052$) and palatal ($d = -1.208$) sounds. The children with cleft palate produced significantly more glottal sounds ($z = -2.772$; $p = 0.006$) than the noncleft children (Table 3).

Multiple Wilcoxon rank-sum tests were conducted to evaluate whether differences existed between the cleft and noncleft children for manner of production categories at 39 months of age. The children with cleft palate produced significantly fewer stops ($z = -3.223$; $p = 0.001$), fricatives ($z = -3.398$; $p = <0.001$), affricates ($z = -2.812$; $p = 0.005$), glides ($z = -2.768$; $p = 0.006$), and liquids ($z = -3.856$; $p = <0.001$), compared to the noncleft children. No significant group differences were found for the percentage of nasal sounds produced (Table 4).

Independent t tests were conducted to compare the cleft and noncleft groups for language measures at 39 months. No significant group difference was found for the number of different words in a 150-utterance sample ($t = -1.554$; $df = 64$; $p = 1.125$). Significant differences were found for both MLU in words ($t = -3.110$; $df = 64$; $p = 0.003$), and MLU in morphemes ($t = -3.357$; $df = 64$; $p = 0.001$) in a 150-utterance spontaneous speech/language sample, with the noncleft children producing significantly longer utterances on both measures (Table 5).

Question Two

Chi-square analyses and a Fisher's exact test were run to determine the association between speech status (i.e., speech within normal limits, or speech not within normal limits) and risk factors (i.e., sex, maternal education, number of episodes of OM, resonance status, and therapy status) for children with cleft palate at 39 months. Results of chi-square analyses indicated a statistically significant association between poor

speech outcomes and sex, interestingly, with females performing worse than males. Additional associations were found between maternal education level and speech status. The children whose mothers had less than a bachelor's degree had significantly poorer speech outcomes than the children whose mothers had at least a bachelor's degree. A chi-square analysis also indicated statistically significant associations between resonance ratings and speech status. Children rated as having normal resonance or mild hypernasality had better speech outcomes compared to children rated as having moderate or severe hypernasality. Results of Fisher's exact test showed no significant associations between number of episodes of OM and speech outcomes, as did the final chi-square analysis between therapy enrollment and speech status (Table 6).

Question Three

Descriptive analyses were employed to determine the number of children with cleft palate who fit into each of the four speech/language outcome profiles (Figure 1). A majority of the children (41%) were categorized as normal VP mechanism + delayed speech and/or language (Profile 2). Within that profile, 82% of the children were delayed just in speech, 12% were delayed in both speech and language, and 6% were delayed in language only (Figure 2). The second most common profile (Profile 1) was normal VP mechanism + normal speech and language (33%). The third most common profile was questionable VP mechanism + delayed speech and/or language (Profile 3; 21%). Within that profile, 56% of the children were delayed in speech only, and 44% were delayed in both speech and language (Figure 3). The last profile, questionable VP mechanism + normal speech and language (Profile 4) had the lowest percentage of children (5%).

Question Four

A linear regression was conducted using the LASSO approach to determine which timepoint(s) and speech and language variables were most predictive of speech and language outcomes at 39 months of age (Table 7). All dashes in the table represent predictor variables that were shrunk to zero by the LASSO approach and therefore had no predictive value for the outcome measure of interest.

When the outcome was TCI at 39 months, the following variables were predictive: TCBR and TCI at 9 months/presurgery (-0.450; -0.927, respectively); TCBR at postsurgery (0.496); and TCI (0.629), percentage of correct consonants (0.072), percentage of true stops at 21 months (0.003), and total correct stop production (0.278). When the outcome was the number of stable consonants at 39 months, the following variables were predictive: TCBR at 9 months/presurgery and postsurgery (-1.761; 1.012, respectively); and stable consonants (0.729), percentage of true stops (0.022), and NDW at 21 months (0.034). When the outcome was percentage of correct consonants at 39 months, the following variables were predictive: TCBR at 9 months/presurgery and postsurgery (-7.503; 2.938, respectively); and TCI (1.003), stable consonants (1.391), percentage of true stops (0.116), total correct stop production (0.596), and NDW (0.141) at 21 months. When the outcome measure was total correct stop production at 39 months, the following variables were predictive: TCBR and TCI at 9 months/presurgery (-8.888; 3.315, respectively); TCBR at postsurgery (4.193); and percentage of correct consonants (0.094), percentage of true stops (0.221), and total correct stop production (3.511) at 21 months. When the outcome measure was NDWs, the following variables were predictive: TCI at 9 months/presurgery (-8.278); and TCI (2.349), percentage of true stops (0.425),

total correct stop production (2.344), and NDWs (0.302) at 21 months. Finally, when the outcome measure was MLU in morphemes, the following variables were predictive: percentage of true stops postsurgery (0.091); and TCI (-0.086), stable consonants (-0.333), and percentage of correct consonants (0.020) at 21 months.

Across all outcomes, the predictor variables at 9 months/presurgery (TCBR and TCI) revealed negative correlations suggesting that children with a lower TCBR and TCI at 9 months of age had better speech and language outcomes at 39 months of age. Additionally, the predictors for MLU in morphemes showed negative correlations with the 21-month measures, TCI and stable consonants, suggesting that the lower the size of TCI and number of stable consonants at 21 months, the higher the MLU at 39 months of age. All other correlations were positive suggesting that better performance on the predictor variables at a younger age were associated with better performance on the speech and language outcome measures at 39 months. It should be noted that for all outcomes with a negative correlation for TCBR or TCI at 9 months/presurgery, the correlation shifted to a positive correlation at postsurgery and 21 months.

Question Five

A logistic regression was conducted using selected factors based on the LASSO approach to determine which timepoint(s) and speech and language variables were most predictive of the four speech/language outcome profiles at 39 months. The outcome profiles were collapsed into 3 categories by combining the normal VP mechanism + delayed speech and/or language group (Profile 2) and the questionable VP mechanism + normal speech/language group (Profile 4), due to the limited sample size. The logistic regression included an odds ratio (OR), which was used to quantify the degree of

association between early speech predictors and the four speech/language outcome profiles. When the point estimation of OR was greater than one, it indicated a positive association with the probability of the child being placed into a certain speech/language outcome profile. When the point estimation of OR was less than one, it indicated a negative association, meaning the child was less likely to be placed into the speech/language outcome profile of interest.

When examining the likelihood that a child would be categorized as having a normal VP mechanism + normal speech/language (Profile 1) versus having a questionable VP mechanism or delayed speech and/or language (Profile 2 or 4), TCI at 21 months had protective effects. This means the larger the child's TCI at 21 months, the lower the risk he/she would have either a questionable VP mechanism or delayed speech and/or language (Profile 2 or 4). Similarly, when examining the likelihood that a child would be categorized as having a normal VP mechanism + normal speech/language (Profile 1) versus a questionable VP mechanism + delayed speech and/or language (Profile 3), TCI and percentage of true stops at 21 months had protective effects. Although both measures were found to be significant, TCI at 21 months had the strongest correlation since the OR point estimation was further from one (Table 8).

Table 2

Mean (*M*), Standard Deviation (*SD*), Median (*Med*), Range, Significance Levels (*p*), and Effect Size (*d*) for Speech Outcome Measures of Children with Cleft Palate and Noncleft Peers at 39 Months

	Cleft		Noncleft		<i>p</i>	<i>d</i>
	<i>M</i> (<i>SD</i>)	<i>Med.</i> (<i>Range</i>)	<i>M</i> (<i>SD</i>)	<i>Med.</i> (<i>Range</i>)		
TCI	15.3 (3.1)	16 (5-21)	17.7 (1.7)	18 (14-21)	<0.001W*	
Stable	10.2 (4)	10 (2-17)	13.7 (2.8)	13 (10-21)	<0.001*	-1.014
%CC	58.9 (15.9)	61.6 (14-81.3)	73.8 (7.5)	72.6 (57.9-90.3)	<0.001W*	

Note. TCI = True consonant inventory; Stable = Stable consonants; %CC = Percent correct consonants.

P values marked with "W" denote the use of a Wilcoxon rank-sum test.

**p* < .05

Table 3

Mean (*M*), Standard Deviation (*SD*), Median (Med), Range, Significance Levels (*p*), and Effect Size (*d*) for Place of Production Measures of Children with Cleft Palate and Noncleft Peers at 39 Months

	Cleft		Noncleft		<i>p</i>	<i>d</i>
	<i>M</i> (<i>SD</i>)	Med. (Range)	<i>M</i> (<i>SD</i>)	Med. (Range)		
% Labial	83.9 (16.7)	90.1 (23.3-99.4)	94.5 (4)	95.6 (86.3-100)	0.001W*	
% Dental	11.4 (16.9)	5.3 (0-71.4)	24.4 (21.2)	19.2 (0-80)	0.002W*	
% Alveolar	51.7 (18.2)	52.5 (4.6-83.6)	67.4 (10.7)	68.8 (47-86.3)	<0.001*	-1.052
% Palatal	45.4 (18.9)	41.9 (14-85.1)	66.6 (16.1)	63.5 (35.4-94.6)	<0.001*	-1.208
% Velar	62 (27.9)	69.8 (3.2-97.7)	80.3 (18.4)	82.5 (8.3-100)	0.003W*	
% Glottal	11.6 (11.1)	7.9 (1.8-65.2)	5.7 (2.4)	5.2 (1.2-10.6)	0.006W*	

Note. *P* values marked with “W” denote the use of a Wilcoxon rank-sum test.

**p* < .05

Table 4

Mean (*M*), Standard Deviation (*SD*), Median (Med), and Significance Levels (*p*) for Manner of Production Measures of Children with Cleft Palate and Noncleft Peers at 39 Months

	Cleft		Noncleft		<i>p</i>
	<i>M</i> (<i>SD</i>)	Med. (Range)	<i>M</i> (<i>SD</i>)	Med. (Range)	
% Stops	57.3 (22.1)	63.8 (3.3-86.8)	73.6 (8)	73.9 (61.1-87.7)	0.001W*
% Fricatives	51.1 (17.4)	53 (16-78.7)	66.7 (8.2)	64.4 (56.3-87.7)	<0.001W*
% Affricates	36.3 (32.5)	30.8 (0-101)	59.6 (26.3)	66.7 (0-100)	0.005W*
% Nasals	81.2 (12.9)	84 (34.3-100)	83.9 (10)	84.6 (50.9-96.5)	0.389W
% Glides	89.9 (10.8)	93.1 (46.2-100)	95.7 (5.3)	97.1 (82.4-100)	0.006W*
% Liquids	28.3 (21.3)	21.6 (3.2-78.4)	51.7 (22.2)	52.9 (17.8-89.3)	<0.001W*

Note. *p* values marked with “W” denote the use of a Wilcoxon rank-sum test.

**p* < .05

Table 5

Mean (*M*), Standard Deviation (*SD*), Median (Med), Range, Significance Levels (*p*), and Effect Size (*d*) for Language Outcome Measures of Children with Cleft Palate and Noncleft Peers at 39 Months

	Cleft		Noncleft		<i>p</i>	<i>d</i>
	<i>M</i> (<i>SD</i>)	Med. (Range)	<i>M</i> (<i>SD</i>)	Med. (Range)		
NDW	124.4 (28.6)	124 (54-176)	136.4 (32.2)	143 (54-203)	0.125	-0.394
MLU_w	2.4 (0.5)	2.4 (1.4-3.7)	2.8 (0.5)	2.8 (1.8-4)	0.003*	-0.800
MLU_m	2.5 (0.6)	2.6 (1.4-4)	3.1 (0.6)	3.1 (1.9-4.5)	0.001*	-1.000

Note. NDW = Number of different words; MLU_w = Mean length of utterance in words; MLU_m = Mean length of utterance in morphemes.

**p* < .05

Table 6

Percentage Totals (%), Number of Participants (*N*), Significance Levels (*p*), and Test Statistics (χ^2) for Speech Status Based on Risk Factors of Children with Cleft Palate at 39 Months

	Speech WNL % (<i>N</i>)	Speech NWNL % (<i>N</i>)	<i>p</i>	χ^2
Sex				
Female	16.7 (3/16)	52 (13/16)	0.018*	5.592
Male	83.3 (15/27)	48 (12/27)		
MomEd				
> Bachelor	55.6 (10/16)	24 (6/16)	0.035*	4.460
< Bachelor	44.4 (8/27)	76 (19/27)		
OM Status				
0 Episodes	11.1 (2/2)	0 (0/2)	0.156F	3.926
1+ Episodes	50 (9/19)	40 (10/19)		
Chronic	38.9 (7/22)	60 (15/22)		
Resonance				
Normal	72.2 (13/21)	32 (8/21)	0.009*	6.776
Hypernasal	27.8 (5/22)	68 (17/22)		
Therapy				
Yes	61.1 (11/27)	64 (16/27)	0.847	0.037
No	38.9 (7/16)	36 (9/16)		

Note. WNL = Speech within normal limits; NWNL = Speech not within normal limits; MomEd = Maternal education level; > Bachelor = At least a bachelor's degree; < Bachelor = Less than a bachelor's degree; OM status = number of episodes of otitis media; 1+ Episode = 1 to 4 episodes of OM; Chronic = 5 or more episodes of OM; Normal = resonance rating of normal or mild hypernasality; Hypernasal = resonance rating of moderate or severe.

p value marked with "F" denote the use of Fisher's exact test. All other associations were chi-squares.

**p* < .05

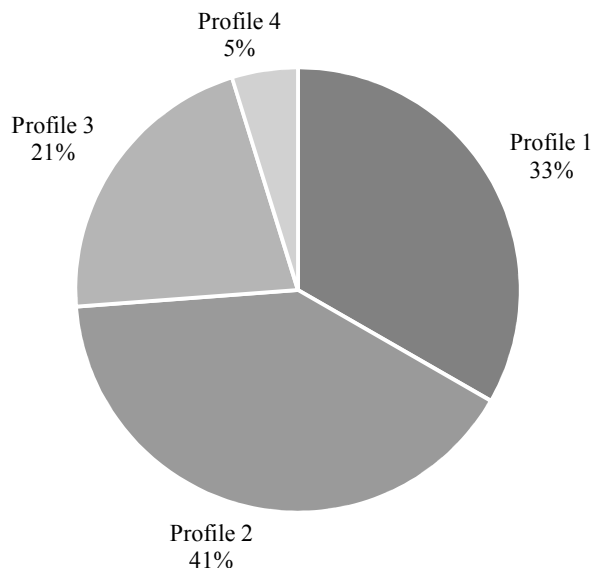


Figure 1. Percentage of children with cleft palate in each of the four speech/language outcome profiles. Profile 1 = Normal VP mechanism + normal speech and language; Profile 2 = Normal VP mechanism + delayed speech and/or language; Profile 3 = Questionable VP mechanism + delayed speech and/or language; Profile 4 = Questionable VP mechanism + normal speech and language.

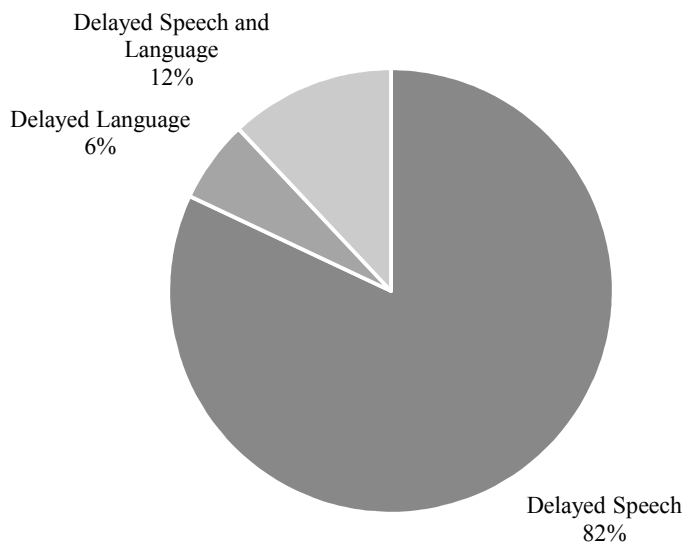


Figure 2. Percentage of children with delayed speech, delayed language, or delayed speech and language for Profile 2, normal VP mechanism + delayed speech and/or language.

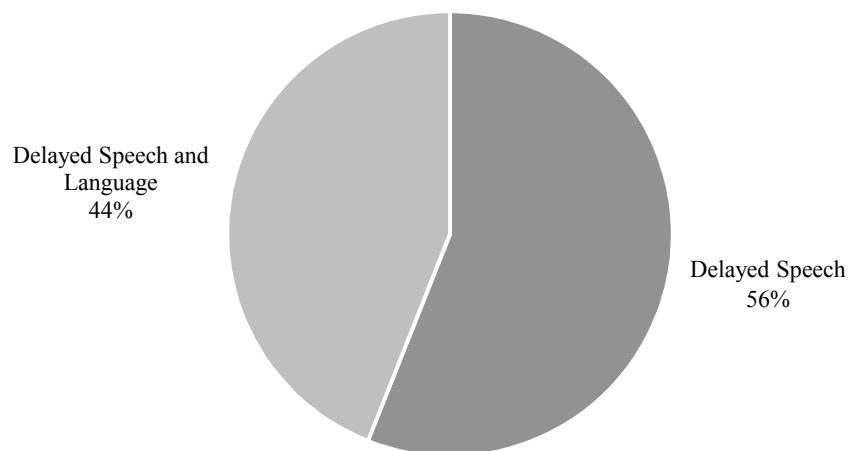


Figure 3. Percentage of children with delayed speech or delayed speech and language for Profile 3, questionable VP mechanism + delayed speech and/or language.

Table 7

Estimate of Coefficients and Standard Errors for Speech and Language
Outcome Measures Based on Early Predictors for
Children with Cleft Palate

	39 Month Outcomes					
	TCI	Estimate of Coefficients (Standard Errors)			NDW	MLUm
		Stable	%CC	TC Stops		
9 TCBR	-0.450 (0.507)	-1.761 (0.506)	-7.503 (2.330)	-8.888 (3.621)	-	-
9 TCI	-0.927 (0.513)	-	-	-3.315 (3.695)	-8.278 (4.158)	-
Post TCBR	0.496 (0.524)	1.012 (0.596)	2.938 (2.772)	4.193 (3.780)	-	-
Post %True Stops	-	-	-	-	-	0.091 (0.129)
21 TCI	0.629 (0.498)	-	1.003 (2.968)	-	2.349 (5.126)	-0.086 (0.106)
21 Stable	-	0.729 (0.547)	1.391 (3.260)	-	-	-0.333 (0.137)
21 %CC	0.072 (0.043)	-	-	0.094 (0.301)	-	0.020 (0.010)
21 %True Stops	0.002 (0.030)	0.022 (0.030)	0.116 (0.163)	0.221 (0.208)	0.425 (0.276)	-
21 TC Stops	0.278 (0.737)	-	0.596 (4.204)	3.511 (5.062)	2.344 (5.594)	-
21 NDW	-	0.034 (0.030)	0.141 (0.152)	-	0.302 (0.263)	-

Note. TCBR = True canonical babbling ratio; TCI = True consonant inventory; Stable = Number of stable consonants; %CC = Percentage of correct consonants; % True Stops = Percentage of true stops; NDW = Number of different words; MLUm = Mean length of utterance in morphemes; TCStops = Total correct stop production.

9 = 9 months/presurgery; Post = Postsurgery; 21 = 21 Months of age

Table 8

Odds Ratio Point Estimations and 95% Confidence Intervals (*CI*), and Coefficient Estimation and Standard Error (*SE*) for Speech Outcome Profiles Based on Early Predictors in Children with Cleft Palate at 39 Months

	Point Estimation	Odds Ratio (<i>CI</i>)	Coefficient Estimation	(<i>SE</i>)
Profile 1 vs. Profile 2 or 4				
21 TCI	0.763	(0.585, 0.996)	-0.270	(0.136)
Profile 1 vs. Profile 3				
21 TCI	0.767	(0.534, 1.103)	-0.265	(0.185)
21 % True Stops	0.952	(0.858, 1.057)	-0.049	(0.053)
21 % Total Correct Stops	0.967	(0.897, 1.043)	-0.033	(0.039)

Note. Profile 1 = Normal VP mechanism + normal speech and language, Profile 2 = Normal VP mechanism + delayed speech and/or language, Profile 3 = Questionable VP mechanism + delayed speech and/or language, Profile 4 = Questionable VP mechanism + normal speech and language; TCI = True consonant inventory; % True stops = Percentage of true stops; % Total correct stops = Total correct stop production. 21 = 21 Months of age

DISCUSSION

This study addressed multiple questions related to speech and language of 3-year-old children with cleft palate. First, the study compared the speech and language of children with cleft palate with their noncleft peers at 39 months. Second, the study examined risk factors for delayed speech and language skills in children with cleft palate at 39 months of age. Third, the study identified the number of children with cleft palate who fell into each of the four speech/language outcome profiles: 1) normal VP mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, 4) questionable VP mechanism + normal speech and/or language (Scherer et al., 2005). Finally, the study determined which age and speech/language measures were most predictive of later speech outcomes for children with cleft palate at 39 months of age.

Speech Skills of Children with Cleft Palate Compared to Noncleft Peers

The children with cleft palate in the current study were behind their noncleft peers for a majority of the speech and language measures of interest. Starting with size of TCI, the children with cleft palate had approximately two fewer consonants in their inventory compared to noncleft children of the same age. This was similar to the findings of Jones et al. (2003) showing that 17-month-old children with cleft palate had a TCI size of approximately three fewer consonants than a comparison group. Other studies looking at younger children with cleft palate ranging from 13 months to 27 months found that they

produced between two to four fewer consonants than their noncleft peers; however, those studies did not examine true consonant usage specifically (Chapman & Hardin-Jones, 1992; Hardin-Jones & Chapman, 2014; Scherer et al., 2008). Rather, they compared size of consonant inventory which should show less separation between the groups as sounds included in the size of consonant inventory count, but excluded from the TCI count, are more commonly occurring in the speech of children with cleft palate (e.g., glides and glottals) (Chapman et al., 2001; Scherer et al., 2008). Additionally, in the present study, the children with cleft palate had approximately three fewer stable consonants compared to the noncleft children, and their percentage of consonants correct differed by approximately 15% (CP (M) = 59%; NC (M) = 74%). No other studies using a noncleft group for comparison have analyzed stable consonants. It should be noted however that there was more variability in the range of stable consonants for the children with cleft palate (range = 2-17) than the noncleft children (range = 10-21). Scherer et al. (2008) examined percentage of consonants correct at 30 months and found the children with cleft palate were about 16% lower than same age peers, which was consistent with the present findings.

Results indicated that place of production characteristics showed consistencies and inconsistencies with previous studies. In the present study, the children with cleft palate produced significantly fewer place features with the exception of glottals. As early as 9 months of age, Chapman et al., (2001) found significant group differences in the use of velars and glottals, and nonsignificant differences for all other place features. Jones et al. (2003) found significant differences in the use of alveolars only at age 17 months. The findings by Chapman et al. (2003) comparing 21-month-old children from the same data

set as the present study were the most relevant for comparison because their findings also examined the child's accuracy in words, whereas the previous studies examined the various place features in prelinguistic productions primarily. Chapman and colleagues found statistically significant differences between the cleft and noncleft groups for all but two place features. One explanation for this apparent widening of the gap between cleft and noncleft groups is that as the children get older, they are attempting a greater variety of words, and therefore sounds. As they attempt more words and sounds, the sounds they are targeting are also later developing and thus more difficult to produce. Another explanation is that some of the other earlier studies had fewer participants (as few as 14 per group in the Jones et al. [2003] study), so they may not have been adequately powered to detect differences. However, none of these studies provided effect sizes as part of their results.

The findings of this study indicated that as a group, children with cleft palate differed from their noncleft peers in almost all manner of production characteristics at 39 months as well. The children with cleft palate produced significantly fewer stops, fricatives, affricates, glides and liquids. Scherer et al. (2008) compared the production of consonant sounds between 30-month-old children with cleft palate and their noncleft peers. They found fewer of the children with cleft palate produced stops, fricatives, affricates, and liquids in comparison to noncleft children. Chapman et al., (2003) found statistically significant differences for 21-month-old children with and without cleft palate for production of stops and liquids. Historically, stops, fricatives, affricates, and liquids have been found to be less frequently occurring and/or less accurate in the speech of children with cleft palate at varying ages (Chapman et al. 2001; Chapman & Hardin,

1992; Jones et al., 2003). While the explanation for less accurate production of pressure consonants (e.g., stops, fricatives, affricates) is likely related to early or current VPD, liquid production should not be impacted by a cleft palate. However, it has been shown that children with cleft palate and VPD were more likely to misarticulate these sounds, probably due to a more general speech delay (see Peterson-Falzone, 2010 for a review).

Results of the language analysis revealed no significant group differences for the number of different words in a 150-utterance spontaneous speech/language sample. However, most studies of younger children with cleft palate found they have lower expressive vocabularies compared to noncleft peers (Broen et al., 1998; Hardin-Jones & Chapman, 2014; Jones et al., 2003), with some studies suggesting the vocabulary gap widens as they age (Broen et al., 1998; Hardin-Jones & Chapman, 2014). Many of the previous studies used parent report measures to assess expressive vocabulary skills as the children were younger (Broen et al., 1998; Hardin-Jones & Chapman, 2014), whereas the present study calculated the number of different words from a spontaneous speech/language sample while the child was interacting with their primary caregiver. A study by Collett and colleagues (2010) found that 5-year-old children with cleft palate were not delayed compared to peers on standardized tests of receptive and expressive vocabulary skills. It is possible that vocabulary size of the participants in this study was somewhat inflated as the speech and language samples included a combination of spontaneous and imitated words (Miller & Chapman, 2000). Or it might be that expressive vocabulary is an area of strength for children with cleft palate. Clearly, more data, preferably longitudinal data, is needed to clarify this issue.

Statistically significant group differences were noted for both MLU in words and

MLU in morphemes. The differences found for MLU were consistent with previous studies of cleft and noncleft children (Morris, 1962; Scherer & D'Antonio, 1995; Spriesterbach et al., 1958). However, not all studies have found that children with cleft palate exhibit lower MLUs (Broen et al., 1998; Jocelyn et al., 1996;). Some researchers have questioned whether lower MLUs in children with cleft palate are related to language deficit per se or reflect the child's efforts to improve intelligibility by reducing utterance length (Faircloth & Faircloth, 1972; Morris, 1968; Scherer & D'Antonio, 1995). Chapman, Hardin-Jones, Moreau, and Fetrow (2015) found correlations between intelligibility and MLU for children with cleft palate but not for noncleft children, supporting this hypothesis. Many studies have advocated for the use of MLU as a general measure of expressive language in young children (Brown, 1973; Dethorne, Johnson, & Loeb, 2004; Miller, 1981; Nice, 1925; Rice et al., 2010). However, when using MLU to identify language delay in children with cleft palate, speech skills should also be considered.

Predictors of Speech Outcomes in Children with Cleft Palate at 39 Months of Age

The next analysis identified the important risk factors (e.g., sex, maternal education, history of otitis media, etc.) associated with poor speech outcomes for children with cleft palate at 39 months. Of the variables examined, sex, maternal education, and resonance status were associated with delayed speech at 39 months. The most surprising finding was that female sex was associated with poorer speech outcomes when compared to males. This was inconsistent with what had been previously reported in the literature for children with and without cleft palate, as we know that more males than females

required secondary surgery (Bicknell, McFadden, & Curran, 2002), and male sex is a risk factor for speech delay for children in the general population (Campbell et al., 2003). Hardin-Jones, Brown, Van Demark, and Morris (1993) looked at children ages 4 to 16 and found that females with cleft palate had greater rates of change than males which was associated with a better speech outcome. Since the children in that study were older, it is unknown whether the females had better speech at younger ages, or if they had worse speech which therefore led them to make more gains as they aged. Riski and Delong (1984) examined children with cleft palate aged 3 to 8 years and found no significant differences in articulation performance based on sex.

In order to clarify the impact of sex on speech outcomes at 39 months, a post hoc analysis was carried out using percentage of correct consonants rather than speech status, as determined by the Smit et al. (1990) norms, as the speech outcome variable. No significant differences were found between males and females for percentage of correct consonants. Based on the Smit et al. (1990) norms, females acquired one more consonant than males by 39 months, and that consonant was /d/. This likely accounted for the lower performance of females in this study since children with cleft palate are known to have additional difficulties with pressure consonants, specifically stops. So, while more females were judged to be delayed by the Smit et al. normative data, they did not differ from boys in the number of sounds in error.

A poorer speech outcome was also associated with a lower level of maternal education. This finding is consistent with a study of risk factors in a large cohort of 3-year-old children with speech sound delays but without cleft palate. Campbell et al. (2003) found that children of mothers who had not completed high school were at the

greatest risk for later speech delays. For children with cleft palate, Yun et al. (2016) found that parental education level was one of the most significant risk factors for speech delays for children with cleft palate living in China. Maternal education has been suggested as a measure of socioeconomic status because of its association with income, healthcare, environment, cognition, and language development (Fujiura & Yamaki, 2000; Satcher, 1995; Siegel, 1982; Smith, Brooks-Gunn, & Klebanov, 1997; Zill, 1996). Children of mothers with less education may live in a less stimulating environment and have less access to resources such as speech therapy, healthcare, and preschool services (Smith et al., 1997).

Findings related to the impact of maternal education in the current study are particularly compelling as the groups were based on whether or not the mother had a bachelor's degree or less. Other studies have found that maternal education of less than high school is the most predictive category (Campbell et al., 2003; Dollaghan et al., 1999). We were not able to make these distinctions in level of education due to sample size constraints.

The findings of a relationship between resonance status and speech status seem reasonable as children with moderate to severe hypernasality exhibit VPD which interferes with the production of high pressure consonants. Although nasal air emission and weak pressure were not counted as errors in the current study, clearly this group exhibited more speech sound errors in general. The strong relationship between resonance and speech production can be seen in these data as only 5% of the children fell into the profile of questionable VP function + normal speech/language compared to 21% being classified as questionable VP function + delayed speech and/or language

development.

One unexpected finding was the lack of an association between the number of episodes of OM and speech outcome at 39 months. Although many studies have tried, it is difficult to isolate the impact of OM and associated hearing loss on speech and language functioning of children with cleft palate. Until better methodologies are developed to document OM and hearing in status in children with cleft palate, we can only speculate about the impact of chronic OM on speech and language outcomes for these children (Hardin-Jones & Chapman, 2014).

Speech Outcome Profiles

This was the first study to look at speech outcomes in terms of profiles; therefore, limited comparisons could be made with previous studies. The first profile included children with normal VP mechanism and normal speech/language. The present study found 33% of children fell into this profile. Of the children with normal VP function and normal speech/language (Profile 1), 58% had been enrolled in therapy and 42% were never enrolled in therapy. This suggested that only 12% were able to achieve normal speech status without speech and/or language intervention. This was considerably lower than the 50% that was proposed by Spriestersbach et al. (1973). This finding highlights the impact of the cleft on early development. The children in this study had primary palatal surgery at a median age of 12 months, which is consistent with what is typically seen across most centers in the United States and Canada (Chapman, personal communication, February 2017; Katzel et al., 2009). Some have advocated for earlier surgery, at approximately 6 months of age and prior to the onset of canonical babbling to provide the baby with a normal speech production mechanism prior to the onset of

canonical babbling (e.g., Chapman et al., 2001; Chapman et al., 2008; Kemp-Fincham, Kuehn, & Trost-Cardamone, 1990). A clinical trial of timing of primary palatal surgery is currently being carried out in Europe to address this question.

The next speech/language outcome profiles included children with delayed speech and/or language and normal VP function (Profile 2; 41%) and delayed speech and/or language and questionable VP function (Profile 3; 21%). Added together, 62% of children with cleft palate in the present study exhibited speech/language delays at 39 months, regardless of VP status. Although this was the first study to report the occurrence of speech/language delay in a consecutive sample of children at age 39 months, these findings were consistent with those of Hardin-Jones and Jones (2005), who found that 68% of the preschoolers they studied (age 2;10 to 5;6) were enrolled in speech/language therapy. This was four times greater than the occurrence of speech delay (15.6%) (Campbell et al., 2003) or six times greater than the occurrence of language delay (8 – 10%) (Dale, Price, Bishop, & Plomin, 2003; Silva, 1980) for 3-year-olds in the general population.

The last factor considered in the speech/language outcome profiles was VP status. This included children falling into Profile 3, questionable VP function and delayed speech and/or language (21%), and Profile 4, questionable VP function and normal speech/language (5%). Across the two profiles, the present study found a total of 26% of children with cleft palate with a questionable VP mechanism at age 39 months. This was somewhat lower than the studies by Hardin-Jones and Jones (2005) and Chapman et al. (2008), who found 37% and 43% of preschool aged children had moderate to severe hypernasality or had secondary surgery for VPD, respectively. These numbers were

substantially higher than reported here as these data did not take into account children who later received secondary surgery. However, when we included children who later had secondary surgery (beyond 39 months of age) and those who were rated as exhibiting moderate to severe hypernasality at 39 months, the number jumped from 26% to 52%.

In determining the four speech/language outcome profiles, we did not consider the data on secondary surgery as the speech/language outcome profiles were meant to be classified based only on the perceptual characteristics observed at 39 months of age. If in fact, all of these children with moderate to severe hypernasality eventually required surgery for VPD, this rate of 52% is high based on what has been reported in the literature. Research studies report as few as 5% to as many as 43% of children requiring an additional surgical intervention for VPD post primary palatal repair (Chapman et al., 2008; Enderby & Emerson, 1995; Lithovius et al., 2014). This variation may be related to a number of variables such as lack of standardized procedures for assessing and rating speech of children with cleft palate, difficulty of perceptual ratings of hypernasality, and different standards across cleft palate teams and/or SLPs about what constitutes clinically significant hypernasality (see Chapman et al., 2016 for a review).

In an effort to understand the discrepancy between the number of children rated as exhibiting a questionable VP mechanism and those who eventually had secondary surgery, we examined the ratings of those children who eventually had secondary surgery and found that 53% of those children who were rated by K. Chapman and M. Hardin-Jones as exhibiting mild hypernasality eventually underwent secondary surgery. What is not clear is whether the hypernasality increased with age or if the high rate of secondary surgery reflected the philosophy of the Cleft Palate-Craniofacial teams who were

managing these children. What we do know, however, is that there is variability across SLPs in their judgments about which children should be referred for evaluation of VP functions even after consensus training for rating this variable (Chapman et al., 2016). It is likely that at least some of the discrepancy is related to differences in decisions about what constitutes clinically significant hypernasality (i.e., hypernasality is deviant enough to warrant surgical intervention).

Variables that Predict Outcomes

39 Month Speech and Language Outcome Variables

Speech-language pathologists who evaluate and provide therapy to children with cleft palate are interested in identifying which variables predict speech outcomes for these children as they age. Not only does this guide assessment practices, but helps to identify “red flags” for children needing early intervention. Further, for children with a questionable VP mechanism following primary palatal surgery, it is important to determine as soon as possible if additional surgery is needed to improve speech intelligibility and overall communicative competence. The identification of these predictors will help us do that.

When examining which age and speech measures were most predictive of later speech outcomes for children at 39 months of age, several patterns emerged. First, TCBR at 9 months/presurgery and postsurgery was predictive of all speech outcome variables at 39 months. Interestingly, however, the 9-month/presurgery TCBR scores were negatively correlated with the outcome variables, but by postsurgery, the scores were positively correlated. These findings were consistent with those of Chapman (2004) and Scherer et al. (2008) who found significant negative correlations between TCBR presurgery (or in

the case of Scherer and colleagues-mean babbling level) and speech outcomes at 39 months, and positive but insignificant correlations between TCBR postsurgery and the 39 months' speech outcome variables. Scherer et al. (2008) attempted to explain these findings by stating that children who babble less presurgery have more gains to make postsurgery compared to children who babble more frequently. To support this theory, Scherer et al. (2008) compared the inventories of the group who babbled less frequently to the group who babbled more frequently. They found the group who babbled less presurgery gained almost double the number of consonants by 30 months (postsurgery) compared to the group who babbled more frequently presurgery. As an alternative explanation, Chapman (2004) suggested that the relationship between early/presurgery performance for children with cleft palate was impacted by interventions that occur in the postsurgery period including surgical intervention as well as speech/language intervention. So, babies who appeared to be doing well presurgery, but had VPD post-palatal surgery could not maintain their high level of performance. Conversely, those babies who were poor babblers presurgery and had a good surgical outcome may eventually catch up on their own or show improvements due to intervention.

By 21 months, the speech variables most predictive of better speech outcomes were related to stop production (percent true stops and total correct stop production). In general, these variables were predictive of the majority of the outcome measures of speech and language at 39 months. One of the most significant findings of the study by Chapman et al. (2003) was the strong correlations between true stop production and later speech and language performance at 21 months. These findings were not as strong in the subsequent study by Chapman (2004) but percentage of true stop production postsurgery

was correlated with language outcomes at 39 months. Normative data by Smit et al. (1990) show that the consonant inventories of typically developing children were loaded with stops, and that children master stop consonants early in development. Peterson-Falzone et al. (2010) describe how children with cleft palate are at a disadvantage because they cannot build up enough intraoral pressure to produce stop consonants (or other pressure consonants) prior to palatal surgery. Lack of growth in stop production postsurgery was one the most important clinical finding for determining which children required early intervention services (Hardin-Jones & Chapman, 2008). Further, poor stop production stops by 21 months and especially 39 months of age would be a sign of possible VPD.

TCI scores at 9 months/presurgery and 21 months were identified as predictors although 21 months had the strongest predictive value, as it was associated with a majority of the speech outcomes. In all but one of the outcome measures, TCI had a positive correlation meaning the more true consonants the child had in their inventory, the better their speech was at 39 months. According to Vihman and Miller (1988) and Thal et al. (1996), this finding is significant because true consonant production appears to be related to the shift from the prelinguistic stage to word productions.

Speech/Language Outcome Profiles

The final question examined the predictor variables in relation to the four speech/language outcome profiles: 1) normal VP mechanism + normal speech and language, 2) normal VP mechanism + delayed speech and/or language, 3) questionable VP mechanism + delayed speech and/or language, 4) questionable VP mechanism + normal speech and/or language (Scherer et al., 2005). These findings confirmed the

strength of TCI and percentage of true stops as predictive values at 21 months. A higher TCI at 21 months was the best indicator of children showing normal speech and resonance (Profile 1) at 39 months compared to the other three profiles. In addition, the percentage of true stops at 21 months was also predictive of normal speech and resonance compared to Profile 3, which was delayed speech/language and questionable VP mechanism.

We can therefore surmise that measures obtained prior to palatal surgery may not be indicative of later speech outcomes. Assessments conducted postsurgery have the advantage that if primary palatal surgery is successful, speech and language development may proceed normally after a period of catch up or following a period of speech/language intervention.

Limitations of the Study

There are several limitations of the present study. The first is that these data were part of a larger longitudinal study that was previously completed; therefore, modifications to the collection of background information, spontaneous speech/language samples, and data analysis were not possible. Although it is a rich data set and still relevant as timing of surgery in the United States has changed little since these data were collected, changes to the initial data collection protocol were not possible. For example, ideally, family history of speech and language delays would have been reported and considered as a predictor factor for speech outcomes at 39 months.

The second limitation of the present study was that, although a standard set of toys were provided and the caregivers were instructed to interact naturally with their children during the spontaneous speech/language samples, some samples included a

larger number of questions and single word imitations as the initial focus of the study was on collection of speech production data rather than language data. This may have resulted in less conversational output from the child, but a larger range of sounds and words attempted, making the samples over or under representative of the child's actual speech and language abilities. Fortunately, this was the case for both the children with cleft palate and their noncleft peers.

The third limitation of the present study was related to the information about OM and hearing status of the children. Although history of OM and hearing status was assumed to be an important risk factor, this information can be difficult to obtain. Information was collected from a number of sources including medical records, parents at the regularly scheduled visits, and physicians who completed data forms when children received medical treatment for OM. In addition, tympanometry was carried out at all study visits, and yearly audiometric evaluations were conducted, and yet, the reliability of the data was still problematic.

Although perhaps not limitations of the study, two additional issues warrant consideration when interpreting the findings. First, the impact of enrollment in this study on the management of the children with cleft palate should be considered. These children were followed closely by the primary investigators of the initial longitudinal study and a research assistant visited the families in their homes at 3-month intervals over the study period. They received testing and feedback about their child's speech, language, and hearing from age 6 months to age 39 months. This may have resulted in a larger number of children receiving aggressive management (i.e., earlier speech therapy, frequent hearing evaluations, etc.) than the general population of children with cleft palate. In

addition, although we reported the number of children who had secondary surgery for VPD after the conclusion of the longitudinal study, the primary investigators were not able to follow up with all of the participants. Therefore, it is unknown whether all of the children who were rated as having moderate or severe hypernasality received a secondary surgery.

Finally, the criteria that were used to determine if the child had a language delay and/or speech delay were determined specifically for this study. A change in the criteria employed would also result in a change in the classification status for a number of the children. The method employed was constrained by the tests that had been administered when the study was designed and there were children who showed variable performance on the three measures. Although a majority were clearly classified as language delayed on all three expressive language measures, not all were. Further, we used the criteria of one standard deviation below the mean to signify a delay on the language measures which may have resulted in a higher percentage of children being classified as language delayed.

Clinical Implications

These findings have important clinical implications for the management of children with cleft palate. First, when evaluating the language skills of a child with cleft palate, speech production accuracy and intelligibility should be taken into account. Second, a child who is not producing true stops by 3 to 6 months postsurgery and certainly by 21 months of age is at risk for speech and language delays and should be enrolled in speech therapy (Chapman et al., 2003). Third, improvements in speech should begin by 3 to 6 months postsurgery, but 21 months of age appears to be the most reliable

age to identify risk factors associated with poorer outcomes for children with cleft palate.

Finally, the speech/language outcome profiles should be used as guidelines for management (Scherer et al., 2003). Children in profile one: Normal VP mechanism + normal speech and language, should be monitored every 3 to 6 months to ensure they are keeping up with noncleft peers. Children in profile two: Normal VP mechanism + delayed speech and/or language, should receive speech therapy focused on eliminating compensatory articulation errors, producing more front sounds, and expanding the child's consonant inventory. Children in profile three: Questionable VP mechanism + delayed speech and/or language, should receive trial therapy to teach correct placement of pressure consonants so VP function can be adequately assessed. Other goals could focus on eliminating compensatory articulation errors, expanding consonant inventory, and increasing vocabulary. Children in profile four: questionable VP mechanism + normal speech and language, may need initial therapy to teach correct placement of pressure consonants, then should be referred for a full assessment of VP function (Chapman, 2016b; Scherer et al., 2005).

Conclusions

The findings of this study indicated that children with cleft palate continued to fall behind their noncleft peers at 39 months for the majority of the speech and language variables assessed. Maternal education level, resonance status, and sex were identified as risk factors of poorer speech outcomes at 39 months. The majority of the children with cleft palate were categorized as having normal VP mechanism + delayed speech/language which suggests there is a need for children with cleft palate to receive earlier speech and language intervention to help them catch up with their noncleft peers. The second most

common profile was normal VP mechanism + normal speech/language. This may not be representative of all populations of children with cleft palate due to differences in management, but it does indicate that some children with cleft palate develop speech and language typically while others are able to catch up with the help of speech therapy. Strong correlations were found between TCI and stop production at age 21 months, and positive speech and language outcomes at 39 months suggesting that 21 months is the best age for predicting later speech and language performance. Also, early speech therapy for children with cleft palate should focus on increasing the child's consonant inventory and production of stop consonants. Future studies should look at additional timepoints postsurgery to determine if other predictors stand out and if the current findings hold up at older ages. Also, a more in-depth study examining the differences between children who develop normal speech and language naturally versus children who catch up with speech therapy may provide further insight into optimal environments and the most beneficial treatment strategies when working with children with cleft palate.

APPENDIX

True Canonical Babbling Ratio (TCBR)

The TCBR is similar to the Canonical Babbling Ratio (CBR) developed by Oller, Levine, Cobo-Lewis, Eilers, and Pearson (1998) in which the total number of syllables is divided by the number of canonical syllables. The TCBR is calculated the same way, except only syllables containing true consonants are counted. A true consonant, as defined by Stoel-Gammon (1989), is any consonant (e.g., stop, fricative, affricate, nasal, liquid) except a glottal or glide (e.g., [h], [ʔ], [w], [j]), excluding any laryngeal or pharyngeal consonants (Chapman, Hardin-Jones, Schulte, & Halter, 2001).

Size of True Consonant Inventory (TCI)

The TCI was derived by counting the number of true consonants that occurred at least two times in two different utterances during the speech and language sample.

Stable Consonants

Consonants were considered stable if they occurred with 70% or more accuracy in the all positions at 39 months, and in initial position only at 21 months.

Percent Accuracy of Consonants in Different Place and
Manner Categories

The percent accuracy was calculated in LIPP (Oller, 1990) by dividing the number of correct occurrences of a specific place or manner category by the total number of opportunities for that same place or manner category.

Percent Correct Consonants

The percentage of correct consonants was calculated using LIPP (Oller, 1990). The number of consonants produced correctly was divided by the number of opportunities for consonant productions.

Percentage of True Stops

The percentage of true stops was calculated using LIPP (Oller, 1990). The number of true stops produced was divided by the total number of consonants produced in the sample.

REFERENCES

- Bayley, N. (1993). *Bayley Scales of Infant Development*. San Antonio, TX: The Psychological Corporation.
- Bernhardt, B.H., Doan, A., & Stoel-Gammon, C. (1995). *Phonological and phonetic analysis of cleft palate speech*. Paper presented at the proceedings of the XIIth International Congress of Phonetic Science, Stockholm.
- Bicknell, S., McFadden, L.R., & Curran, J.B. (2002). Frequency of pharyngoplasty after primary repair of cleft palate. *Journal of Canadian Dental Association*, 68(11), 688-692.
- Blakeley, R.W., & Brockman, J.H. (1995). Normal speech and hearing by age 5 as a goal for children with cleft palate. *American Journal of Speech-Language Pathology*, 4, 25-32.
- Broen, P. A., Devers, M. C., Doyle, S. S., Prouty, J. M., & Moller, K. T. (1998). Acquisition of linguistic and cognitive skills by children with cleft palate. *Journal of Speech, Language, Hearing Research*, 41, 676-687.
- Brown, R. (1973). *A first language: The early stages*. Cambridge, MA: Harvard University Press.
- Bzoch, K. R. (1965). Articulation proficiency and error patterns of preschool cleft palate and normal children. *The Cleft Palate Journal*, 2, 340-349.
- Bzoch, K. R. (1997a). Clinical assessment, evaluation, and management of 11 categorical aspects of cleft palate speech disorders. In K. Bzoch (Ed.), *Communicative disorders related to cleft lip and palate* (pp. 261-311). Austin, TX: Pro-Ed.
- Bzoch, K. R. (1997b). Rationale, methods and techniques of cleft palate speech therapy. In K. Bzoch (Ed.), *Communicative disorders related to cleft lip and palate* (pp. 227-260). Austin, TX: Pro-Ed.
- Campbell, T.F., Dollaghan, C.A., Rockette, H.E., Paradise, J.L., Feldman, H.M., Shriberg, L.D., Sabo, D.L., & Kurs-Lasky, M. (2003). Risk factors for speech delay of unknown origin in 3-year-old children. *Child Development*, 74(2), 346-357.

- Chapman, K.L. (1991). Vocalizations of toddlers with cleft lip and palate. *The Cleft Palate-Craniofacial Journal*, 28(2), 172-178.
- Chapman, K. L. (1993). Phonologic processes in children with cleft palate. *The Cleft Palate-Craniofacial Journal*, 30(1), 64-72.
- Chapman, K. L. (2004). Is presurgery and early postsurgery performance related to speech and language outcomes at 3 years of age for children with cleft palate? *Clinical Linguistics & Phonetics*, 18(4-5), 235-257.
- Chapman, K. L. (2008). Speech and language of children with cleft palate: Interactions and influences. In K. T. Moller & L. E. Glaze (Eds.), *Cleft palate: Interdisciplinary issues and treatment – For clinicians by clinicians* (pp. 243-272). Austin, TX: Pro-Ed Publications.
- Chapman, K. L. (2016). *Principles of intervention* [PDF document]. Class handout.
- Chapman, K. L., Baylis, A. L., Trost-Cardamone, J., Cordero, K., Dixon, A. J., Dobbeltsteyn, C., et al. (2016). The Americleft Speech Project: A training and reliability study. *Cleft Palate-Craniofacial Journal*, 53, 93-108.
- Chapman, K.L., & Hardin, M.A. (1992). Phonetic and phonologic skills of two-year-olds with cleft palate. *Cleft Palate-Craniofacial Journal*, 29(5), 435-443.
- Chapman, K.L., Hardin-Jones, M.A., Goldstein, J.A., Halter, K.A., Havlik, R.J., & Schulte, J. (2008). Timing of palatal surgery and speech outcome. *Cleft Palate Craniofacial Journal*, 45(3), 297-308.
- Chapman, K.L., Hardin-Jones, M., & Halter, K.A. (2003). The relationship between early speech and later speech and language performance for children with cleft lip and palate. *Clinical Linguistic & Phonetics*, 17(3), 173-197.
- Chapman, K.L., Hardin-Jones, M., Moreau, K., & Fetrow, R. (2015). *Language skills of preschoolers with cleft palate*. Poster presented at the American Cleft Palate-Craniofacial Association Annual Conference, Palm Springs, CA.
- Chapman, K.L., Hardin-Jones, M., Schulte, J., & Halter, K.A. (2001). Vocal development of 9-month-old babies with cleft palate. *Journal of Speech, Language, and Hearing Research*, 44, 1268-1283.
- Chapman, K.L., Tecco, G.K., Gooch, J., & Visconti, C. (1998). Conversational skills of preschool and school-aged children with cleft lip and palate. *Cleft Palate-Craniofacial Journal*, 35, 503-516.
- Chapman, K. L., & Willadsen, E. (2011). The development of speech production in children with cleft palate. In S. Howard & A. Lohmander (Eds.), *Cleft lip and*

- palate: Speech assessment, analysis and intervention* (pp. 23-36). Hoboken, NJ: Wiley Publishers.
- Collett, B.R., Leroux, B., & Speltz, M.L. (2010). Language and early reading among children with orofacial clefts. *Cleft Palate-Craniofacial Journal*, 47(3), 284-287.
- Dale, P.S., Price, T.S., Bishop, D.V.M., & Plomin, R. (2003). Outcomes of early language delay: I. Predicting persistent and transient language difficulties at 3 and 4 years. *Journal of Speech, Language, and Hearing Research*, 46, 544-560.
- Dethorne, L., Johnson, B., & Loeb, J. (2005). A closer look at MLU: What does it really measure? *Clinical Linguistics and Phonetics*, 19(8), 635-648.
- Dollaghan, C. A., Campbell, T. F., Paradise, J. L., Feldman, H. M., Janosky, J. E., Pitcairn, D. N., et al. (1999). Maternal education and measures of early speech and language. *Journal of Speech, Language, and Hearing Research*, 42, 1432-1443.
- Dorf, D.S., & Curtain, J.W. (1981). Early cleft palate repair and speech outcome. *Plastic and Reconstructive Surgery*, 70(1), 74-81.
- Enderby, P., & Emerson, J. (1995). *Does speech and language therapy work? A review of the literature*. San Diego, CA: Singular Pub. Group.
- Estrem, T., & Broen, P. A. (1989). Early speech production of children with cleft palate. *Journal of Speech and Hearing Research*, 32, 12-23.
- Faircloth M., & Faircloth S. (1972). Delayed language and linguistic variations. In K. Bzoch (Ed.), *Communicative disorders related to cleft lip and palate*. Boston, MA: Little Brown, 130-135.
- Fenson, L., Dale, P.S., Resznick, J.S., Thal, D.A., Bates, E., Hartung, J.P., et al. (1993). *MacArthur Communicative Development Inventories*. San Diego, CA: Singular Publishing Group, Inc.
- Fey, M. E. (1986). *Language intervention with young children*. San Diego, CA: College-Hill Press.
- Fletcher, S.G. (1978). *Diagnosing speech disorders from cleft palate*. New York: Grune and Stratton.
- Fox, D., Lynch, J., & Brookshire, B. (1978). Selected developmental factors of cleft palate children between two and thirty-three months of age. *Cleft Palate Journal*, 15(3), 239-245.
- Frederickson, M.S., Chapman, K.L., & Hardin-Jones, M. (2006). Conversational skills of

- children with cleft lip and palate: A replication and extension. *Cleft Palate-Craniofacial Journal*, 43(2), 179-188.
- Fujiura, G. T., & Yamaki, K. (2000). Trends in demography of childhood poverty and disability. *Exceptional Children*, 66, 187-199.
- Golding-Kushner, K. J. (2001). *Therapy techniques for cleft palate speech and related disorders*. San Diego, CA: Singular.
- Goldman, R., & Fristoe, M. (1986). *Goldman Fristoe test of articulation*. Circle Pines, MN: American Guidance Service.
- Grunwell, P., & Russell, J. (1987). Vocalisations before and after cleft palate surgery: A pilot study. *International Journal of Language & Communication Disorders*, 22(1), 1-17.
- Grunwell, P., & Russell, J. (1988). Phonological development in children with cleft lip and palate. *Clinical Linguistic & Phonetics*, 2(2), 75-95.
- Hardin-Jones, M.J., Brown, C.K., Van Demark, D.R., & Morris, H.L. (1993). Long-term speech results of cleft palate patients with primary palatoplasty. *Cleft Palate-Craniofacial Journal*, 30(1), 55-63.
- Hardin-Jones, M., & Chapman, K.L. (2008). The impact of early intervention of speech and lexical development for toddlers with cleft palate: a retrospective look at outcome. *Language, Speech, and Hearing Services in Schools*, 39, 89-96.
- Hardin-Jones, M., & Chapman, K.L. (2014). Early lexical characteristics of toddlers with cleft lip and palate. *The Cleft Palate-Craniofacial Journal*, 51(6), 622-631.
- Hardin-Jones, M. A., & Jones, D. L. (2005). Speech production of preschoolers with cleft palate. *Cleft Palate-Craniofacial Journal*, 42(1), 7-13.
- Harding, A., & Grunwell, P. (1996). Characteristics of cleft palate speech. *European Journal of Disorders of Communication*, 31, 331-357.
- Harding, A., & Grunwell, P. (1998). Active versus passive cleft-type speech characteristics. *International Journal of Language & Communication Disorders*, 33(3), 329-352.
- Hodson, B.W., Chin, L., Redmond, B., & Simpson, R. (1983). Phonological evaluation and remediation of speech deviations of a child with a repaired cleft palate: A case study. *Journal of Speech and Hearing Disorders*, 48(1), 93-98.
- Hutters B., Bau A., & Brondsted K. (2001). A longitudinal group study of speech development in Danish children born with and without cleft lip and palate.

- International Journal of Language & Communication Disorders*, 36, 447–470.
- Hutters, B., & Brøndsted, K. (1987) Strategies in cleft palate speech—with special reference to Danish. *Cleft Palate-Craniofacial Journal*, 24, 126–136.
- International Phonetic Alphabet. (1989). International Phonetic Association. Victoria, British Columbia, Canada.
- Jocelyn, L.J., Penko, M.A., & Rode, H.L. (1996). Cognition, communication, and hearing in young children with cleft lip and palate and in control children: A longitudinal study. *Pediatrics*, 97(4), 529-534.
- Jones, C.E., Chapman, K.L., & Hardin-Jones, M.A. (2003). Speech development of children with cleft palate before and after palatal surgery. *Cleft Palate-Craniofacial Journal*, 40(1), 19-31.
- Karnell, M.P., & Van Demark, D.R. (1986). Longitudinal speech performance in patients with cleft palate: Comparisons based on secondary management. *Cleft Palate Journal*, 23(4), 278-288.
- Katzel, E. B., Basile, P., Koltz, P. F., Marcus, J.F., & Giroto, J. A. (2009). Current surgical practices in cleft care: Cleft palate repair techniques and postoperative care. *Plastic and Reconstructive Surgery*, 124(3), 899-906.
- Kemp-Fincham, S. I., Kuehn, D. P., & Trost-Cardamone, J. E. (1990). Speech development and the timing of primary palatoplasty. In J. Bardach & H. L. Morris (Eds.), *Multidisciplinary management of cleft lip and palate* (pp. 736–746). Philadelphia: W.B. Saunders.
- Lithovius, R. H., Ylikontiola, L. P., & Sandor, G. K. (2014). Frequency of pharyngoplasty after primary repair of cleft palate in northern Finland. *Oral Surgery Oral Medicine Oral Pathology Oral Radiology*, 117(4), 430-434.
- Lohmander-Agerskov, A., Soderpalm E., & Friede, H. (1995). A longitudinal study of the speech in 15 cleft lip and palate children with late hard palate repair. *Scandinavian Journal of Plastic and Reconstructive Surgery, Hand Surgery*, 29, 21-31.
- Lohmander-Agerskov, A., Soderpalm, E., & Friede, H. (1996). Delayed hard palate repair: A comparison of speech in children with open and functionally closed residual clefts. *Scandinavian Journal of Plastic and Reconstructive Surgery, Hand Surgery*, 30, 121-127.
- Lohmander-Agerskov, A., Soderpalm, E., Friede, H., Persson, E.C., & Lilja, J. (1994). Pre-speech in children with cleft lip and palate or cleft palate only: Phonetic analysis related to morphologic and functional factors. *Cleft Palate-Craniofacial*

- Journal*, 31(4), 271-279.
- Lohmander, A., Olsson, M., & Flynn, T. (2011). Early consonant production in Swedish infants with and without unilateral cleft lip and palate and two-stage palatal repair. *The Cleft Palate-Craniofacial Journal*, 48(3), 271-285.
- Lohmander, A., Persson, C., & Owman-Moll, P. (2002). Unoperated clefts in the hard palate speech deficits at age 5 and 7 years and the relationship to cleft size. *Scandinavian Journal of Plastic and Reconstructive Surgery, Hand Surgery*, 36, 332-339.
- Long, N.V., & Dalston, R.M. (1982). Gestural communication in twelve-month-old cleft lip and palate children. *Cleft Palate Journal*, 19(1), 57-61.
- Long, N.V., & Dalston, R.M. (1983). Comprehension abilities of one-year-old infants with cleft lip and palate. *Cleft Palate Journal*, 20(4), 303-306.
- Lu, Z., Ma, L., Luo, Y., & Fletcher, P. (2010). The effects of unrepaired cleft palate on early language development in Chinese infants. *Cleft Palate Craniofacial Journal*, 47(4), 400-404.
- Lynch, J., Fox, D., & Brookshire, B. (1983). Phonological proficiency of two cleft palate toddlers with school-age follow-up. *Journal of Speech and Hearing Disorders*, 48(3), 274-285.
- Lynch, M.P., Oller, D.K., & Steffens, M.L. (1989). Development of speech-like vocalizations in a child with congenital absence of cochleas: The case of total deafness. *Applied Psycholinguistics*, 10, 315-333.
- McWilliams, B. J., Morris, H. L., & Sheldon, R. L. (1990). *Cleft palate speech* (2nd ed.). Toronto, Ontario, Canada: B.C. Decker.
- Miller, J. (1981). *Assessing language production in children: Experimental procedures*. Baltimore, MD: University Park Press.
- Miller, J., & Chapman, R. S. (2000). *Systematic Analysis of Language Transcripts: Version 6.1a (SALT)*, [Computer Software]. Madison, WI: University of Wisconsin.
- Morris, H.L (1962). Communication skills of children with cleft lip and palate. *Journal of Speech and Hearing Research*, 5, 79-90.
- Morris, H., & Ozanne, A. (2003). Phonetic, phonological, and language skills of children with a cleft palate. *Cleft Palate-Craniofacial Journal*, 40(5), 460-470.
- Nation, J.E. (1970). Vocabulary comprehension and usage of preschool cleft palate and

- normal children. *Cleft Palate Journal*, 7, 639-644.
- Neiman, G.S., & Savage, H.E. (1997). Development of infants and toddlers with clefts from birth to three years of age. *Cleft Palate-Craniofacial Journal*, 34(3), 218-225.
- Nice, M. (1925). Length of sentences as a criterion of a child's progress in speech. *Journal of Educational Psychology*, 16(6), 370-379.
- O'Gara, M.M., & Logemann, J.A. (1988). Phonetic analyses of the speech development of babies with cleft palate. *The Cleft Palate Journal*, 25(2), 122-134.
- O'Gara, M.M., Logemann, J.A., & Rademaker, A.W. (1994). Phonetic features by babies with unilateral cleft lip and palate. *American Cleft Palate-Craniofacial Association*, 31(6), 446-451.
- Oller, D.K. (1986). Metaphonology and infant vocalizations. In B. Lindblom & R. Zetterstrom (Eds.), *Precursors of early speech* (pp. 21-35). New York: Stockholm Press.
- Oller, D.K. (1990). Logical International Phonetic Program [Computer Software]. Miami: Intelligent Hearing Systems.
- Oller, D.K. (2000). *The emergence of the speech capacity*. Mahwah, NJ: Lawrence Erlbaum Associates.
- Oller, D.K., Levine, S.L., Cobo-Lewis, A.B., Eilers, R.E., & Pearson, B.Z. (1998). Vocal precursors to linguistic communication: How babbling is connected to meaningful speech. In R. Paul (Ed.), *Exploring the speech-language connection* (pp. 1-23). Baltimore, MD: Paul H. Brookes.
- Oller, D.K., & Lynch, M.P. (1992). Infant vocalizations and innovations in infraphonology: Toward a broader theory of development and disorders. In C. Ferguson & L. Menn & C. Stoel-Gammon (Eds.), *Phonological development: Models, research, implications* (pp. 509-539). Parkton, MD: York Press.
- Oller, D.K., Wieman, L.A., Doyle, W.J., & Ross, C. (1976). Infant babbling and speech. *Journal of Child Language*, 3, 1-11.
- Pamplona M.C., & Ysunza, A. (2000). Active participation of mothers during speech therapy improved language development of children with cleft palate. *Scandinavian Journal Plastic and Reconstructive Surgery, Hand Surgery*, 34, 231-236.
- Pamplona, M.C., Ysunza, A., & Espinosa, J. (1999). A comparative trial of two modalities of speech intervention for compensatory articulation in cleft palate

- children, phonologic approach versus articulatory approach. *International Journal of Pediatric Otorhinolaryngology*, 49, 21–26.
- Pamploma, M.C., Ysunza, A., & Ramirez, P. (2004). Naturalistic intervention in cleft palate children. *International Journal of Pediatric Otorhinolaryngology*, 68(1), 75-81.
- Paradise, J.L., Bluestone, C.D., & Felder, H. (1969). The universality of otitis media in 50 infants with cleft palate. *Pediatrics* 44, 35-42.
- Parent Report Measures. FIRST WORDS Project. (2007). The Florida State University Research Foundation. Available at: <http://firstwords.fsu.edu/parentsReportMeasures.html#>. Assessed October 10, 2010.
- Peterson-Falzone, S. J. (1995). Speech outcomes in adolescents with cleft lip and palate. *Cleft Palate-Craniofacial Journal*, 32(2), 125-128.
- Peterson-Falzone, S., & Graham, M.S. (1990). Phoneme-specific nasal emission in children with and without physical anomalies of the velopharyngeal mechanism. *Journal of Speech and Hearing Disorders*, 55, 132-139.
- Peterson-Falzone, S. J., Hardin-Jones, M. A., & Karnell, M. P. (2010). *Cleft palate speech* (4th ed.). St. Louis, MO: Mosby.
- Peterson-Falzone, S.J., Trost-Cardamone J.E., Karnell, M.P., & Hardin-Jones, M.A. (2006). *The clinician's guide to treating cleft palate speech*. St Louis, MO: Mosby.
- Philips, B. J., & Harrison, R. J. (1969). Articulation patterns of preschool cleft palate children. *The Cleft Palate Journal*, 6, 245-253.
- Philips, B. J., & Harrison, R. J. (1969b). Language skills of preschool cleft palate children. *The Cleft Palate Journal*, 6(2), 108-119.
- Powers, G., Dunn, C., & Erikson, C. (1990). Speech analysis of four children with repaired cleft palates. *Journal of Speech and Hearing Disorders*, 55, 542-550.
- Priester, G. H., & Goorhuis-Brouwer, S. M. (2008). Speech and language development in toddlers with and without cleft palate. *International Journal of Pediatric Otorhinolaryngology*, 72, 801-806.
- Rice, M., Smolik, F., Perpich, D., Thompson, T., Rytting, N., & Blossom, M. (2010). Mean length of utterance levels in 6-month intervals for children 3 to 9 years with and without language impairments. *Journal of Speech, Language, and Hearing Research*, 53(2), 333-349.

- Riski, J. (1979). Articulation skills and oral-nasal resonance in children with pharyngeal flaps. *Cleft Palate Journal*, 16(4), 421-428.
- Riski, J.E. (1995). Speech assessment of adolescents. *Cleft Palate-Craniofacial Journal*, 32(2), 109-113.
- Riski, J.E., & DeLong, E. (1984). Articulation development in children with cleft lip/palate. *Cleft Palate Journal*, 21,57-64.
- Rynnel-Dagoo, B., Lindberg, K., Bagger-Sjoback, D., & Larson, O. (1992). Middle ear disease in cleft palate children at three years of age. *International Journal of Pediatric Otorhinolaryngology*, 23, 201-209.
- Salas-Provance, M.B., Kuehn, D.P., & Marsh, J.L. (2003). Phonetic repertoire and syllable characteristics of 15-month-old babies with cleft palate. *Journal of Phonetics*, 31, 23-38.
- Satcher, D. (1995). Annotation: The sociodemographic correlates of mental retardation. *American Journal of Public Health*, 85, 304–306.
- Scherer, N., Chapman, K. L., Hardin-Jones, M., & D’Antonio, L. (2005). *Early assessment and intervention for children with cleft palate*. American Speech-Language-Hearing Association Convention. San Diego, CA. Invited program participant. Sponsored by Special Interest Division 5 – Speech Science and Craniofacial Disorders.
- Scherer, N.J., & D’Antonio, L. (1995). Parent questionnaire for screening early language development in children with cleft palate. *Cleft Palate-Craniofacial Journal*, 32, 7-13.
- Scherer, N. J., Williams, A. L., & Proctor-Williams, K. (2008). Early and later vocalization skills in children with and without cleft palate. *International Journal of Pediatric Otorhinolaryngology*, 72, 827-840.
- Sell, D., & Ma, L. (1996). A model of practice for the management of velopharyngeal dysfunction. *British Journal of Oral & Maxillofacial Surgery*, 34, 357-363.
- Sell, D., Grunwell, P., Mildinhall, S., Murphy, T., Cornish, T., Bearn, D., et al. (2001). Cleft lip and palate care in the United Kingdom – The clinicial standards advisory group (CSAG) study. Part 3: Speech Outcomes. *American Cleft Palate-Craniofacial Association*, 38(1), 7-30.
- Shriberg, L. D., Austin, D., Lewis, B. A., McSweeny, J. L., & Wilson, D. L. (1997). The Percentage of Consonants Correct (PCC) metric: Extensions and reliability data. *Journal of Speech, Language, and Hearing Research*, 40(4), 708-722.

- Shriberg, L. D., & Kwiatkowski, J. (1994). Developmental phonological disorders I: A clinical profile. *Journal of Speech and Hearing Research, 37*, 1100–1126
- Shulman, B.B. (1985) *Test of pragmatic skills, revised*. Tucson, AZ: Communication Skill Builders.
- Siegel, L. (1982). Reproductive, perinatal, and environmental factors as predictors of the cognitive and language development of preterm and full-term infants. *Child Development, 53*, 963–973.
- Silva, P.A. (1980). The prevalence, stability and significance of developmental language delay in preschool children. *Developmental Medicine and Child Neurology, 22*, 768-777.
- Smit, A., Hand, L., Freilinger, J., Bernthal, J., & Bird, A. (1990). The Iowa articulation norms project and its Nebraska replication. *Journal of Speech and Hearing Disorders, 55*, 779-798.
- Smith, J., Brooks-Gunn, J., & Klebanov, P. (1997). Consequences of living in poverty for young children's cognitive and verbal ability and early school achievement. In G. Duncan & J. Brooks-Gunn (Eds.), *Consequences of growing up poor* (pp. 132–189). New York: Russell Sage Foundation.
- Spiestersbach, D.C., Darley, F.L., & Morris, H.L. (1958). Language skills in children with cleft palates. *Journal of Speech and Hearing Research, 1*(3), 279-285.
- Spiesterbach, D.C., Dickson, D., Fraser, F.C., Horowitz, S.L., McWilliams, B.J., Paradise, J.L., & Randell, P. (1973). Clinical research in cleft lip and cleft palate: The state of the art. *Cleft Palate Journal, 10*, 113-165.
- Stoel-Gammon, C. (1989). Prespeech and early speech development of two late talkers. *First Language, 9*, 207-224.
- Stoel-Gammon, C., & Cooper, J. (1984). Patterns of early lexical and phonological development. *Journal of Child Language, 11*, 247-271.
- Stool, S.E., & Randall, P. (1967). Unexpected ear disease in infants with cleft palate. *Cleft Palate Journal, 4*, 99-103.
- Thal, D., Bates, E., Zappia, M.J., & Oroz, M. (1996). Ties between lexical and grammatical development: Evidence from early-talkers. *Journal of Child Language, 23*, 349-368.
- Trost, J.D., (1981). Articulatory additions to the classical description of the speech of persons with cleft palate. *Cleft Palate Journal, 18*, 93–203.

- Van Demark, D.R., Morris, H.L., & VandeHaar, C. (1979). Patterns of articulation abilities in speakers with cleft palate. *Cleft Palate Journal*, *16*, 230-239.
- Vihman, M. M. (1992). Early syllables and the construction of phonology. In C. A. Ferguson, L. Menn, & C. Stoel-Gammon (Eds.), *Phonological development: Models, research, implications* (pp. 393-422). Monkton, MD: York Press.
- Vihman, M. M., Ferguson, C., & Elbert, M. (1986). Phonological development from babbling to speech: Common tendencies and individual differences. *Applied Psycholinguistics*, *1*, 3-40
- Vihman, M. M., & Greenlee, M. (1987). Individual differences in phonological development: Ages one and three years. *Journal of Speech and Hearing Research*, *30*, 503-521.
- Vihman, M.M., Macken, M.A., Miller, R., Simmons, H., & Miller, J. (1985). From babbling to speech: A re-assessment of the continuity issue. *Language*, *61*, 397-445.
- Vihman, M.M., & Miller, R. (1988). Words and babble at the threshold of lexical acquisition. In M.D. Smith & J.L. Locke (Eds.), *The emergent lexicon: The child's development of linguistic vocabulary* (pp. 151-183). New York: Academic Press.
- Warr-Leeper, G., Crone, L., Carruthers, A., & Leeper, H. (1988). *A comparison of the performance of preschool children with cleft lip and/or palate on the Test of Pragmatic Skills*. Presented at the Annual Meeting of the American Cleft Palate Association, Williamsburg, VA.
- Wetherby, A.M., & Prizant, B.M. (1992). *Communication and Symbolic Behavior Scales-Normed Edition*. Austin, TX: Pro-Ed.
- Willadsen, E. (2013). Lexical selectivity in Danish toddlers with cleft palate. *The Cleft Palate-Craniofacial Journal*, *50*(4), 456-465.
- Willadsen, E., & Albrechtsen, H. (2006). Phonetic description of babbling in Danish toddlers born with and without unilateral cleft lip and palate. *The Cleft Palate-Craniofacial Journal*, *43*(2), 189-200.
- Yun, C., Wang, Z., He, P., Guo, C., Chen, G., & Zheng, X. (2016). Prevalence and parental risk factors for speech disability associated with cleft palate in Chinese children – A national survey. *International Journal of Environmental Research and Public Health*, *13*, 1168-1176.
- Zambrana, I.M., Pons, F., Eadie, P., & Ystrom, E. (2014). Trajectories of language delay from age 3 to 5: Persistence, recovery and late onset. *International Journal of*

Language and Communication Disorders, 49(3), 304-316.

Zill, N. (1996). Parental schooling and children's health. *Public Health Reports*, 111, 34-43.

Zimmerman, I.L., Steiner, V.G., & Pond, R.E. (1992). *Preschool Language Scales-3*. San Antonio, TX: The Psychological Corporation.