METAPHONOLOGY, PHONOLOGY AND LANGUAGE IN CHILDREN WITH CLEFT PALATE

by

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B.A., The University of Ottawa, 1998

A THESIS SUBMITTED IN PARTIAL FULFILLMENT OF THE

REQUIREMENTS FOR THE DEGREE OF

MASTER OF SCIENCE

in

THE FACULTY OF GRADUATE STUDIES

(School of Audiology and Speech Sciences)

We accept this thesis as conforming

to the required standard

THE UNIVERSITY OF BRITISH COLUMBIA

September, 2000

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ABSTRACT

The purpose of this study was to examine the metaphonology and possible associated factors in children with non-syndromic cleft lip and palate. This prospective study involved 20 children who presented with non-syndromic cleft lip and cleft palate only. The 8 males and 12 females ranged in age from 66 to 84 months. Standardized tests were administered to assess the following abilities: metaphonology, phonology, morpho-syntax and vocabulary, verbal working memory and non-verbal cognition. The results indicated that these children perform within normal limits on tests of metaphonology and on tests of morpho-syntax and vocabulary. Ninety percent of the children did have residual speech production problems appear to be related to dental issues such as malocclusions and missing teeth.

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ACKNOWLEDGMENTS

I would like to thank B.B. a.k.a. "Bicycle Barbara" for picking me up and guiding me on this journey.

I would also like to thank Judith Johnston for her constant support and encouragement.

Thank-you to Lynn Tissington for sparking my initial interest in working with children with cleft palate and for encouraging me to do research in this area.

Additional thanks go to Carolyn Johnson for lending me her lap-top when mine crashed.

To my friends Carole, Michelle, "Island" Steph, "Illinois" Steph, and Hilary; thank-you for making me take breaks.

I would also like to thank my dad for editing the orthodontic section and for our Thursday morning chats, and my mom for helping me to stuff and address recruitment letters after the marathon. To my sisters Tamara and Emily, thank-you for all the encouraging chats.

Financial support for this project was provided by the British Columbia Medical Foundation and by a UBC HSS grant.

DEDICATION

This work is dedicated to children with cleft palate and their families throughout British Columbia.

This work is also dedicated to my late grandparents who inspired me to work hard and to follow my dreams.

CHAPTER ONE

INTRODUCTION

The present study was motivated for several reasons. The original reason was in response to Richman, Eliason and Lindegren's (1988) work which reported a higher than average prevalence rate of reading disorders among children with cleft palate. Several questions remained unanswered by this study. The first question is whether there are underlying speech processing difficulties contributing to these deficits. There is a body of literature which has documented the link between deficits in phonological awareness and later reading difficulties (See Catts, 1996, for a review). Unfortunately the metaphonological abilities of this population have not been documented. Information regarding verbal working memory and phonological awareness performance will contribute to the literature on phonological processing abilities in this population.

The Richman et al. (1988) study raised further questions regarding cleft-type differences. For the most part, the body of literature outlining language development in children with cleft palate fails to report cleft-type differences. In addition subject selection methods have not always excluded children who have syndromes associated with cognitive delay. Given the recent advances in medical genetics, it is now possible to detect more of these syndromes than was previously possible (Shprintzen & Goldberg, 1995). Therefore this study was motivated to improve the subject selection process and to report the language abilities of children without known syndromes.

In order to understand the development of speech and language abilities of children with cleft palate it is necessary to first understand the theories associated with language learning in general. Thus, this chapter will begin with a brief outline of language learning theories. The next section will review general literature (including language learning) in the population of interest, that is, the cleft palate population. The final section of this chapter will return to the main focus of the paper in language learning: acquisition of phonological awareness skills, as a background for the research with the cleft palate population.

Theoretical perspectives in speech production

The development of speech and language abilities of children with cleft palate has been traditionally viewed from a medical perspective. Historically, the speech production difficulties of children with cleft palate were considered the result of a congenital abnormality of the oral structure. With recent advances in the area of clinical phonology, there is evidence to suggest that a subgroup of children may have "phonological impairment," in addition to articulatory difficulties (Chapman, 1993; Chapman & Hardin, 1992).

In order to evaluate the theoretical soundness of this new evidence, one must first have an understanding of the four theoretical perspectives which have been used to explain speech and language impairment. The following discussion is based largely on Stackhouse and Well's (1997) review of the classification of speech production problems. An attempt will be made where possible, to relate these perspectives to the development of speech and language abilities of children with cleft palate.

The medical perspective has been adopted as the primary framework in which to view the development of speech and language abilities of children with cleft palate. The medical model provides the diagnosis of the type of clefting condition (bilateral or unilateral cleft lip and palate, cleft palate only) and establishes the theory that the structural deficiencies in the oral mechanism and hearing apparatus are the primary causes of speech difficulties in this population (Stackhouse, 1982; Stackhouse and Wells, 1997). Advances in medical genetics have led to an increased identification of the number of children who have syndromes associated with cognitive delay and subsequent speech and language delay (Shprintzen & Goldberg, 1995). There continue to be cases however, in which the medical model fails to explain why two children with bilateral cleft lip and palate may present with a different profile of speech and language problems. In order to better

understand the varying speech and language profiles, one must consider evidence from the linguistic approach.

Followers of one linguistic tradition, such as Jakobson in the 1940's and Chomsky in the 1960's, proposed a generative view of language learning. They argued that infants possess a highly enriched mental structure which predisposes them to language acquisition. Speech language pathologists frequently use the following linguistic terms to classify delay in various domains: phonology, syntax, semantics and pragmatics. In speech-language pathology terms, phonological delay refers to the child's inability to use sounds to convey meaning. The work by Chapman (1993) suggests that children with cleft palate may have early phonological problems because the loss of phonemic contrastivity renders their speech unintelligible to the listener. In addition to proposing a hierarchical structure to language, the generative linguistic approach views language as a domain separate from other types of cognition. This approach has been widely criticized by psycholinguists who argue that language arises from the interaction of several general cognitive processes.

The psycholinguistic approach was developed in the late 1960's in an attempt to integrate the theories from the fields of linguistics and psychology. This approach generates models of speech processing in an attempt to explain both normal and disordered speech output. The processing models can be empirically tested to establish the level of breakdown which results in disordered speech output. Concepts such as phonological awareness and phonological working memory are used in this approach. Stackhouse and Wells (1997) note that it is "important to distinguish a phonological problem - a term used to describe children's speech output difficulties in a linguistic sense - from a phonological processing problem- a psycholinguistic term used to refer to the specific underlying cognitive deficits that may give rise to speech and/or literacy difficulties (pg. 8)."

In an attempt to examine the complex relationship between articulation and phonology in children with cleft palate, Howard (1993) cites Hewlett's (1990) 'two-lexicon' theory of phonological processing and phonetic production. This psycholinguistic

model posits an input and an output lexicon. The input lexicon consists of single words which are presented in terms of their auditory-perceptual features. In turn these features are mapped by realizational rules to the 'output lexicon' which consists of articulatory representations. This model suggests that it is possible for a word to be correctly represented in the input lexicon, but articulatory constraints such as a faulty velopharyngeal mechanism may alter the phonological representations in the output lexicon. In addition, this model highlights the difference between a speaker-oriented and listener-oriented perspective, i.e., intended vs. actual productions. Howard notes that a detailed phonetic transcription allows you to determine whether the speaker may be signaling contrasts in such a way that the listener cannot perceive them. If such contrasts exist, then this is evidence that the speaker does not have problems at the organizational phonological level; rather, they have implementation problems which reduce the ability to signal phonological contrasts to the listener. In her case study of a child with cleft palate, Howard concluded that the child had an "articulatory disorder which has phonological implications for the listener" (pg. 314). In a recent article Hewlett, Gibbon and Cohen-McKenzie (1998) revised the model and removed the articulatory based 'output lexicon' on the basis that it cannot explain non-word production. The new model has an input lexicon which reflects the "current state of the child's phonological perception" (pg. 170). The output lexicon is now associated with perception because the phonological string is "delivered for production and does not contain any articulatory instructions" (pg. 171). Hewlett et al. (1998) suggest that "articulatory routines are learned through trial and error as the child attempts to replicate the perceptual properties of the input pronunciation using the information provided by auditory memory" (pg. 171). Similar models to that of Hewlett et al. (1998) have been proposed by Stackhouse and Wells (1997). In light of this new model, further study is warranted to determine whether children with and without cleft palate establish robust phonological representations at the perceptual level. In addition, further documentation of the phonological processing abilities of children is needed in

order to gain a better understanding of nature of developmental speech and language deficits, for children with and without speech disorders. While psycholinguistic models provide an account for the components involved in mapping between form and meaning and the nature of processing within and between components, they do not allow for testing real time processing.

In recent years considerable attention has been paid to interactive activation models which have been used to explain language processing. While models vary in terms of their architecture (unidirectional vs. bi-directional, feed forward vs. feed backward etc.) and algorithms (back propagation or changing connection weights etc.), the general premise is that there is activation/inhibition within and among language "units." Unlike modular-linguistic theory, the language units of syntax, lexicon, phonology and phonological rules are not 'encapsulated' or considered to be 'domain specific.' Instead, language acquisition occurs as the units interact. Bates and Goodman (1997) provide evidence which supports the argument that there is a "powerful link between grammar and lexical growth" (pg. 524). They suggest that language acquisition occurs in a non-linear fashion and conclude that "the kinds of critical mass effects that we have proposed to underlie the relation between lexical and grammatical growth may be a special case of this more general approach to the non-linear dynamics of learning" (pg. 544). From this perspective phonological and morphosyntactic disorders can co-occur because there is an interdependence among language units. Phonological and morphosyntactic deficits may lead to inappropriate feedback within the network. As a result, reduced activation levels will inhibit lexical or phonemic retrieval. This model predicts that children with phonological and morphosyntactic deficits may experience difficulties with phonological awareness tasks (and consequently, literacy tasks) because they have weak activation of phonological units. In addition, this model predicts that if children with cleft palate have phonological deficits, they may also be at risk for morphosyntactic deficits. Bernhardt and Stemberger (1998) address the issue of separating articulation from phonology in cleft

palate speakers. Secondary surgery (pharyngoplasty) outcomes appear to shed light on this issue. The authors point to two outcome groups, those whose compensatory articulations resolve after surgery and those who continue to have difficulties. Those with successful post-surgical outcomes may have had an intact underlying phonological system, with "phonetic implementation compromised by the mechanism" (pg. 607). The underlying phonological system of the poor outcome group may be compromised because of an inadequate mechanism and "habitual" patterns.

Future research in the area of cleft palate speech and language development should consider the recent advances from all three perspectives: medical (medical genetics), linguistic (clinical phonology), psycholinguistic (phonological processing) and interactive activation (co-occurrence of phonological and morphosyntactic disorders). This new information should have an impact on subject description and methods of testing and analysis. In turn, it could potentially help to identify certain subgroups of children with cleft palate who are more at risk for speech and language difficulties.

CLEFT PALATE

Cleft Palate: Epidemiology and Genetics

Cleft Palate is the fourth most common birth defect affecting approximately 1 of every 1000 live births (Gorlin et al., 1990). Disruption of embryological development between weeks 6-10 may result in a cleft palate. Clefting can occur as a feature in over 400 genetic syndromes (Shprintzen & Goldberg, 1995) but cleft lip and palate (CLP) and cleft palate only (CPO) can occur in isolation. In terms of speech and language development, the structural defects affect articulators, mechanisms for controlling oronasal resonance, hearing, speech production and voice. It is beyond the scope of this paper to review all the known speech and language issues associated with each clefting condition. Instead the focus will be on hearing, velopharyngeal competency and the concomitant speech production and language issues of children with cleft lip and palate and cleft palate only.

It is important to highlight the differences between cleft lip and palate (CLP) and cleft palate only (CPO) as they are different disorders. Shprintzen and Goldberg (1995) note that the disorders differ in the following ways (1) the lip and the palate are formed from different embryonic elements (2) the lip is formed approximately two weeks before the palate. The authors suggest that the timing aspect alone prevents the two disorders from arising from the same abnormal process.

Recent advances in medical genetics have helped clinicians diagnosis the primary etiology of the cleft. Shprintzen and Goldberg (1995) urge clinicians to advocate for the "child with a cleft" not the "cleft palate child" because it is now known that clefting is not merely a specific disease entity, but rather a possible feature/symptom of a given syndrome.

Cognitive-Developmental Measures

Only a few studies have investigated aspects of early development in children with cleft palate. What follows is a summary of a few studies which document the abilities of infants and preschool children with cleft palate.

In an attempt to expand the literature on the early development of children with cleft lip and palate and cleft palate, Kapp-Simon and Krueckeberg (2000) assessed 180 children using the Bayley Scales of Infant Development (BSID) (Bayley, 1969). This assessment tool has the following subscales: eye-hand coordination, manipulation of items, conceptual abilities, imitation-comprehension and vocalization-social. A cross-sectional analysis was used to examine children's BSID scores across four age groups (6, 12,18 and 24 months); this design was used to replicate an earlier study by increasing the sample size. A longitudinal analysis was used to examine the BSID scores of 85 children after 6 months. At the first testing point, subjects ranged in age from 4 to 15 months, while at the second testing point subjects were 16 to 36 months of age. Subjects were excluded if they had medical problems associated with cognitive delay (e.g. chromosomal abnormality and spina bifida). The results from the cross-sectional and longitudinal data indicated that while infants with Cleft Lip and Palate had scores within the average range across the age groups, over time there was a decrease in scores combined with an increase in standard deviations. The authors were surprised to find that 15.3% of the infants at T2 demonstrated global developmental delay, given that subject selection had excluded children with major syndromes. A stepwise multiple regression analysis revealed that motor/visual perceptual scales accounted for approximately 30% of the variance at T2. Further results indicated that before 15 months of age, the ability to produce and comprehend language did not correlate with global developmental delay at 2 years of age.

Unfortunately no control group of non-cleft peers was used in this study. There is no indication of (a) whether genetic testing was used to determine whether children with cleft palate only had an associated syndrome and (b) the age at which genetic testing occurred. If testing did not take place, then one interpretation of the results could be that the 15% of children who demonstrated developmental delay could have had undiagnosed syndromes which affected their development.

In a similar study, Jocelyn, Penko and Rode (1996) assessed 16 children with unilateral and bilateral cleft lip and palate (CLP) and a control group using the following measures: BSID, Receptive-Expressive Emergent Language Scale (REEL)(Bzoch & League, 1971), Sequenced Inventory of Communication Development (SICD-R)(Hedrick, Prather & Tobin,1984), Preschool language scale (PLS) (Zimmerman, Steiner & Pond, 1979), language sample, visual reinforcement audiology and tympanograms. Subjects were tested at 12 months and again at 24 months. The results of the hearing and language data will be discussed in a later section. The mean BSID scores for the CLP group were within normal limits; however when compared to the scores of the non-cleft group, the children with CLP exhibited significant delays in the development in both motor co-ordination and cognitive ability. The interpretation of the results may be limited due to the small sample size. Despite the small sample, this study contributed to the literature as it controlled for syndromes like Velo-Cardio Facial syndrome by assessing children with CLP, not CPO.

Broen, Devers Doyle, Prouty and Moller (1998) documented the cognitive development of 28 with cleft palate and a control group during their first 30 months. In addition to examining cognitive development, this study investigated the relationship between measures of language, hearing and velopharyngeal function (for which results will be discussed in later sections). Assessments of hearing, middle ear function and language samples were conducted every three months, and cognitive assessments were administered

at 24 and 30 months. The results from the mental scale of the Bayley Scale of Infant Development (BSID) and the Minnesota Child Development Inventory (Ireton & Thwing, 1980) indicated that children with cleft palate performed within normal limits compared to the normative data. However, their results differed significantly from the control group of noncleft children. In particular, the cleft-palate group demonstrated delays in verbal, items but not non-verbal items. They also found a significant difference between groups on rate of vocabulary acquisition. The authors argue that if children with cleft palate differ from the non-cleft group on measures of language performance, (not non-linguistic development) then differences in cognitive skills are language based.

These studies reported that infants and toddlers with cleft palate fall within normal limits on the BSID measure; however recent evidence by Broen et al. (1998) suggest that children with cleft palate differed significantly on measures of language production. Several questions remain unanswered: What are the origins of these delays? To what extent do these delays contribute to the overall development of children with cleft palate? And do these minimal delays resolve themselves over the years?

In an attempt to expand the literature in the area of cognitive development, Eliason and Richman (1990) examined the cognitive abilities of preschoolers with cleft lip and palate. Subjects ranged in age from 4 years to 6 years and none had a hearing loss of greater than 25 dB, a known syndrome or a diagnosis of cognitive delay. The 65 subjects with CLP and CPO were assessed using measures of picture and auditory association, vocabulary, and colour span. As a whole,the preschoolers with cleft palate had normal vocabulary development, and adequate skills on verbal analogies and verbal memory span tasks. The preschool children showed deficits on picture association and color span tasks. The author suggests that these two measures are associated with the active strategy of verbal mediation (verbal rehearsal, labeling and categorization). Results did not indicate a cleft-type distinction.

The research in the area of early cognitive development is scarce. However the results point to deficits in language production and visual motor-perceptual areas for infants and toddlers and verbal mediation in the preschool years. Questions examining the etiologies for these delays remain unanswered and further longitudinal research is needed to determine whether these deficits persist and what, if any, impact they have on subsequent speech and language development.

Hearing

Due to structural and functional abnormalities in the outer, middle, and inner ear, children with cleft palate are at risk for hearing problems. Most commonly, these children experience middle ear effusions as a result of Eustachian tube dysfunction. Paradise (1980) cites the prevalence of otitis media in infants with cleft palate as 100% prior to primary repair. Normal Eustachian tube function plays a key role in ventilating the middle ear; when it is open, it allows for the release of mucous and the equalization of pressure. The tensor palatini muscle plays a role in opening the Eustachian tube. There are several theories which attempt to explain the effect that cleft palate has on Eustachian tube function. Some authors (Witzel, 1995; Stool, 1990) have argued that the inability to dilate the Eustachian tube sufficiently is due to a weak or improper insertion of the tensor veli palatini muscle. Broen, Moller, Carlstrom, Doyle, Devers & Keenan (1996) suggested that prior to primary palatal repair, the tensor muscle fibers fail to follow the normal insertion path into the midline of the palate. The result of this abnormal insertion is that the muscle has insufficient anchorage to open the Eustachian tube properly. Others have suggested that Eustachian tube dysfunction is a result of: abnormal muscles, adenoid hypertrophy and hypercompliance of the Eustachian tube (Bluestone, 1971; Rood and Stool, 1981 as cited in Witzel, 1995). Whatever the cause, children with cleft palate experience difficulty

draining middle ear fluid as a result of poor ventilation, which in turn contributes to the associated mild-moderate conductive hearing loss. The next section will examine the relationship between hearing loss and speech-language development in normals. This discussion serves to provide background information for future sections which will be specific to the issues of cleft palate.

Impact of Otitis Media on Normal Language Development

There has been ongoing debate into the literature regarding the relationship between recurrent otitis media with effusion (OME) and speech and language disorders. One argument is that OME has no long term effects on speech and language development because it causes only temporary mild to moderate hearing loss. This theory contends that any effects may be overcome by normal hearing between episodes of OME and by the fact that after age three there is a decrease in the occurrence of the disease.

Others contend that mild fluctuating hearing loss early in life may interfere with speech perception. The acoustic-phonetic account suggests that hearing loss results in a speech input signal which is degraded and inconsistent. In turn, this may interfere with the ability to make fine phonetic discriminations and to segment speech, thereby impeding the ability to form linguistic categories during a crucial period of language learning.

It is difficult to have a clear understanding of the arguments in this controversy as the previous research in this area is flawed. Studies conducted retrospectively typically report data on the frequency, type and duration of episodes, but not on hearing levels (Shriberg, Friel-Patti, Flipsen & Brown, 2000). Friel-Patti and Finitzo (1990) noted that retrospective studies are confounded by the subject selection process, inaccurate estimates of the number and length of past episodes of OME (usually parental report), and by the lack of double-blind controls. In addition, Shriberg et al. (2000) argue that the measures used to assess speech production abilities may not be sensitive enough for some children

whose problems surface later in development. The results of the Shriberg et al. (2000) study indicated that children with a +20dB hearing loss from 12 to 18 months were more at risk for a subsequent speech disorder (33%) than children with less than 20 dB loss during this period (2%). The authors provide the following two potential explanations of the results: (1) direct effects: acoustic-phonetic account (2) indirect "mediated" effects: cognitive linguistic account.

The acoustic phonetic account claims that the hearing loss associated with otitis media directly affects the perception, discrimination and subsequent representation of phonemes. The indirect account suggests that acoustic-phonetic constraints alone are not sufficient to cause speech and language delay. Rather, speech and language delay is mediated by the interaction of several factors such as genetic factors affecting language acquisition, early recurrent OME, hearing loss, environmental factors. The next section will examine the relationship between OME and phonological development.

OME and Normal Phonological Development

In a recent study Petinou, Swartz and Gravel (1999) examined the relationship between otitis media and early phonological development. A prospective cohort design was implemented; 16 infants children were assessed bi-monthly from 10 months to 14 months. Otoscopic, tympanometric and audiometric assessments were utilized to determine the OME status. Groups were formed on the basis of overall +OME status. Speech samples were collected and the following three measures were used for analysis: rate of vocal output, place of articulation, proportional occurrence for manner of articulation.

There was no difference between +OME and –OME groups on the rate of vocal output measure. The +OME group showed a preference for bilabial stop consonants whereas the –OME group indicated a preference for alveolars and nasals. Children with

otitis media may rely more heavily on visible speech cues because they provide greater feedback, leading to a labial 'default' place of articulation.

In summary, the recent prospective research indicates that there is a relationship between recurrent otitis media and speech and language disorders. It appears that recurrent otitis media, with a hearing loss of 20 dB or greater, interferes with early phonological development and later speech and language development. This research appears to have implications for children with cleft palate as they are at risk for hearing problems related to otitis media. The following sections will (1) examine the OME management issues and (2) outline research which has examined the impact of OME on language development in children with cleft palate.

OME Management of Children with Cleft Palate

It is important to note the difference in management of OME in the cleft and non-cleft populations. Current approaches to OME management range from aggressive to passive. Aggressive management involves the insertion of bilateral myringotomy tubes, while a more moderate approach may be to treat with antibiotics. The most passive and noninvasive method is to simply wait for the middle ear to clear. Insertion of myringotomy tubes allows for pressure equalization of the middle ear space.

Broen et al.(1996) argued that prior to primary palatal repair, middle ear problems in this population should not be considered intermittent. The authors suggested that without surgical intervention, children with cleft palate will have Eustachian tube dysfunction. Therefore OME should be treated differently with the cleft palate population. In their 1996 study, Broen et al. examined the early hearing histories of 28 children with cleft palate and cleft lip and palate as well as a control group of 29 noncleft children. The results indicated that 85% had tubes placed at primary palatal repair (range from 9 to 22 months, mean 13.3 months) while fewer than 30% of the children with cleft lip and palate

had tubes inserted at the time of lip repair. They found that in the cleft palate population, ventilation tubes played a key role in the maintenance of normal middle ear function. Without tubes, middle ear function was not normal and as a result, these children experienced depressed hearing. The authors advocated for early ventilation tube placement, preferably at the time of lip repair.

Impact of Hearing Loss on Language Development in Children with Cleft Palate

In their investigation of early communication abilities of children with cleft lip and palate, Jocelyn et al. (1996) collected data on the hearing status of 16 infants. Visual reinforcement audiology and impedance measures were used to assess hearing status while various measures were used to assess language abilities (REEL,1971; PLS,1979; SICD-R, 1984, and a language sample). The data indicated that the children with CLP had a higher incidence of middle ear disease and more ventilation tubes than the control group. The number of children who had mild to moderate hearing loss at 12 and 24 months was four, a relatively small number of the children with cleft palate (the rest had hearing within normal limits). At 12 months, 31% had ventilation tubes and by 24 months the number had increased to 62%. The authors found a relationship between hearing status at 12 months and production and comprehension language scores at 24 months. While the hearing data were collected prospectively, no information was provided regarding the mean age of initial ventilation tube placement. These results should be interpreted with caution, given that no information regarding the number and duration of otitis media episodes was provided. While the authors suggested that hearing status affected both language comprehension and production skills, they did not mention the mean age of primary palatal repair, nor did they measure articulation abilities, both of which are factors that can contribute to the variability in the language production scores.

In an attempt to account for the differences in language abilities, Broen et al. (1998) collected prospective hearing information. This study investigated the relationship between hearing status, language ability, cognitive ability and velopharyngeal function. Their results indicated that when hearing was added as a covariate, none of the differences previously observed on the linguistic scale of the BSID were significant. The authors therefore argued that hearing loss associated with middle ear effusion can have a negative impact on language development. In addition, the authors pointed to velopharyngeal function as an additional variable which can contribute to a delay in language production.

Phonological/Articulation Skills

In light of recent advances in the field of clinical phonology, there have been debates as to whether the speech errors of children with cleft palate are articulatory and or phonologic in nature (McWilliams, 1990). Most of the previous research has been done from a phonetic viewpoint which assumes that the speech production errors are a result of the structural deficiencies associated with cleft palate. The majority of this research describes the articulation abilities of children with cleft palate as below age-expectations (Fletcher, 1978; Phillips & Harrison, 1969; Riski & Delong, 1984; Van Demark, Morris & Vande Harr, 1979). In these studies, articulation tests were administered and results were described in terms of correct production at the phoneme level and then classified into manner of articulation. All of these studies noted that individual phoneme productions were scored as correct or incorrect, and only Phillips and Harrison (1969) mentioned whether the errors were classified as distortions, substitutions and or omissions. In general, these studies suggested that children with cleft palate are vulnerable to difficulties with fricative and affricate production.

Chapman (1993) suggested that children with cleft palate may also be at risk for phonological disorders. This study examined the phonological processes of 30 pre-school

children with cleft palate and 30 non-cleft children. Subjects were administered the Goldman Fristoe Test of Articulation (GFTA) (Goldman & Fristoe, 1969) and results were analyzed using the Khan-Lewis Phonological Analysis (KLPA) (Khan & Lewis, 1986). The results indicated that the three-year-olds with cleft palate used the following phonological processes more frequently than the control group: final consonant deletion, syllable reduction, backing, stopping, stridency deletion, and deaffrication. The authors noted that these processes are not atypical and are often seen in children without clefts. They also attempt to explain that the process of backing in children with cleft palate may be associated with velopharyngeal incompetency. The authors found that by age 5 children with cleft palate were performing similar to their non-cleft counterparts. The results of this study differ from the articulation results of Riski & Delong (1984) etc. because they suggest that at an early age, children with cleft palate exhibit problems at the organizational phonological level in addition to implementation problems.

More research in the area of phonological processing is needed to ascertain whether children with cleft palate incorporate the early speech sound errors that are associated with structural deviations into their phonological system and whether the phonological problems are indicative of an overall language delay, whether the deviations are strictly articulatory, or whether the difference is possible to determine (as discussed in Howard, 1993).

The following section will address the structural deficits which influence cleft palate speech production. Speech production problems related to velopharyngeal function and the dental and occlusal problems will be discussed.

Velopharyngeal Function

Structural and functional abnormalities of the palate frequently result in an inadequate velopharyngeal mechanism. Structural defects include overt or submucous clefts of the hard and soft palate. Speech production can be affected in children with cleft

palate because the oral and nasal cavities are not separated. Normal velopharyngeal valving is a complex process which involves several velopharyngeal muscles. McWilliams, Morris and Shelton (1990) highlight the research which suggests that the velopharyngeal mechanism does not operate in a binary fashion (i.e., closed for oral speech and open for nasal consonants). The authors note that variables such as vowel height, voicing and proximity to nasal consonants play a role in the degree to which the velopharyngeal mechanism elevates (pg. 203). It is important to note that velopharyngeal dysfunction can stem from several etiologies. Trost-Cardamone (1989) provides a taxonomy which differentiates between velopharyngeal inadequacy, insufficiency, incompetence, and mislearning. Velopharyngeal inadequacy is a general term which encompasses all types of abnormal velopharyngeal function. In contrast, velopharyngeal insufficiency refers to a structural or mechanical interference, while incompetence suggests a neurogenic basis. Velopharyngeal mislearning refers to errors such as phoneme specific nasal emissions which are produced in the absence of structural defects or neurogenic etiology. Velopharyngeal inadequacy can lead to resonance and articulation disorders such as hypernasal resonance, abnormal nasal air emission, weak oral pressure sounds, or compensatory articulatory/phonological errors. The perception of hypernasality occurs when there is an inappropriate synchronicity between movements of the oral and nasal cavities. When this inappropriate coupling of the cavities occurs, sound is directed through the nasal passageways and speech is perceived as hypernasal. Sell, Harding and Grunwell (1992) classify two degrees of hypernasality. Grade one refers to perception on vowels only, while grade two refers to perception on voiced consonants. Hypernasality is not the only form of abnormal resonance. Hyponasality is perceived when normal nasal resonance is reduced due to the blockage of the nasal airway. Blockage can occur for reasons such as an upper respiratory infection or a wide pharyngeal flap. When the blockage occurs in the nasal cavity, cul-de-sac resonance can be perceived. Accurate assessment of resonance

disorders is crucial to the appropriate management of children with repaired palatal clefts. Speech-language pathologists use a variety of instrumental and subjective measures for assessment. To date there is no single standard method for assessing and assigning diagnostic severity classifications to children with repaired palatal clefts who present with resonance disorders (Hirschberg & Van Demark, 1997).

Audible nasal air emissions often occur with production of oral high pressure consonants. If the velopharyngeal port does not obtain adequate closure, the airflow is forced through the nasal passageway and the increased resistance can be perceived as nasal turbulence. In contrast, nasal air emissions can also be inaudible, but visible upon production of stops as measured by using a nasal mirror.

Compensatory articulations are frequently associated with cleft palate speech, occurring in response to velopharyngeal insufficiency. Trost (1981) classified the following types of substitutions: glottal stops, pharyngeal stops, pharyngeal fricatives, mid-dorsum palatal stops, and posterior nasal fricatives. These substitutions are often referred by the term "backing." Velopharyngeal insufficiency prevents the build-up of oral pressure and the place of articulation is moved below the velopharyngeal port (Sell et al., 1992). The substitutions occur when the speaker tries to maintain the manner of articulation of the target phoneme.

Double articulations occur when there are two simultaneous articulations in different places. For example, corarticulation can occur with glottal and oral stops.

Speech and Orthodontic Issues

In addition to producing errors related to velopharyngeal insufficiency, children with cleft palate produce errors which are a result of dental and skeletal malocclusions. It is difficult to predict speech outcomes based on diagnosis alone. Factors such as the severity of the original malformation, timing and nature of primary palatal surgery

(LeBlanc & Cisneros, 1995), and individual differences in the range of normal development for exfoliation and eruption of teeth (Aduss & Figueroa, 1990), should be considered, thus making prognostic statements difficult. Cleft-type differences will be discussed from a speech production perspective. However, given the heterogeneity of development, these are only broad generalizations.

Bilateral cleft lip and palate is marked by the greatest degree of tissue deficiency as the cleft occurs under both nostrils (Subtelny, 1990). At birth the premaxilla is relatively protrusive, and this abnormal position affects the development of dental arches and oral functioning (swallowing, tongue position, speech etc). There are missing teeth in the cleft area, and this group is more prone to fistulae in the anterior portion of oral cavity. In cases of unilateral cleft lip and palate the alveolar ridge is cleft which affects dental and occlusal development.

Le Blanc and Cisneros (1995) discuss the relationship between speech production and dentition. What follows is a summary of their discussion on the relationship between structural deformities and articulation abilities.

Teeth: Missing teeth can distort the production of the alveolar sounds /s/, /z/ and palato-alveolar sounds / ʃ ,tʃ dʒ / the tongue may move forward into the space resulting in a frontal lisp. Alternatively the tongue may retract, resulting in a mid-palatal fricative or lateral fricative.

Occlusion: Malocclusions of the maxillary and mandibular arches can interfere with the following phonemes /t,d,f,v,s,z,ʃ, tʃ, dʒ/. When the maxilla is abnormally positioned, the tongue is prevented from acquiring a normal resting position in the cavity. This has an impact on maxillary development which can later effect speech production. Severe maxillary protrusion can prevent the adequate closure to create a seal for bilabial sounds. In addition labiodental sounds may also be minimally affected. Sibilants and affricates are

also affected as maxillary protrusion can impact the direction of airflow needed for this manner of articulation. Lateral distortions are often associated with maxillary protrusion as lingual protrusion or retraction can co-occur, thus changing the place of articulation and interfering with the grooving aspect of the tongue. In addition grooving of the tongue can be prevented by a constricted maxillary arch. The constricted arch prevents the tongue from coming in contact with the borders of the maxillary teeth, thus leading to lateral distortions. Mandibular protrusion has essentially the same effect on articulation. However sibilant production may be affected in a slightly different fashion. Interdental distortions are often perceived to be a result of lingual protrusion. In addition to maxillary and mandibular malocclusions, open and cross- bites result in lateralized, retroflexed distortions as lingual alveolar contact and airflow dynamics are affected.

Upper airway obstruction due to a deviated septum or small nares due to the nasal deformity, often lead to mouth breathing which causes similar problems with tongue position and inhibits maxillary development.

Timing of Primary Palatal Repair

There has been debate in the literature with regards to the proper timing of initial palatal repair. Speech-language pathologists have traditionally advocated for early surgical intervention because this allows for the functional speech mechanism to be restored, thus preventing the onset of compensatory behaviors. On the other hand, orthodontists have argued for later surgery to ensure that it does not interfere with maxillofacial growth development (Kemp-Finchman, Kuehn, Trost-Cardamone, 1990).

McWilliams et al.(1990) provided a summary of ten studies which indicated that earlier palatal repair (prior to 12 months of age) is related to better speech outcomes. In an attempt to determine the sensitive period for primary surgery, Kemp-Finchman et al. (1990) considered aspects of phonologic, anatomic-physiologic and motor development.

They concluded that the optimal time for surgery was 4 to 6 months of age. Unfortunately this hypothesis has yet to be tested.

Language Skills: Morphosyntax and Vocabulary

While the majority of the research has been focused on the speech production abilities of children with cleft palate, there have been a number of investigations which report on the language abilities of this population (e.g. Broen et al., 1998; Dalston, 1990; Fox, Lynch & Brookshire, 1978; Jocelyn et al., 1996; Lynch, 1986; Nation, 1970; Richman, 1980; Richman & Eliason, 1984 ; Smith & McWilliams, 1968). These studies primarily provide evidence that the language skills of children with cleft palate are delayed compared to their non-cleft peers.

One of the first studies to look beyond the speech production abilities of this population was Smith and McWilliams (1968). The authors used the Illinois Test of Psycholinguistic Abilities (ITPA) (Kirk, McCarthy & Kirk, 1968) to evaluate the language abilities of 136 children with cleft palate. Subjects ranged in age from 3 to 8;11 and included those with cleft lip, cleft lip and palate and cleft palate only. No attempt was made to control for IQ, socio-economic status, hearing loss or syndromes associated with cognitive delay. Children with cleft palate only exhibited language production difficulties and visual memory deficits, while children with cleft lip and palate exhibited a similar profile which was not as marked. The authors expressed their surprise at the finding of general visual-motor deficits. However, this finding could be explained by the presence of a syndrome such as velo cardio facial syndrome.

Language delay in the preschool years has been documented in a few studies. Studies should be interpreted with caution as many fail to adequately control for syndromes associated with cognitive delay and many fail to differentiate between the cleft types,

factors which could account for the delayed performance on language and other developmental measures.

Nation (1970) used the Peabody Picture Vocabulary Test (PPVT) (Dunn, 1959) to assess the vocabulary skills of 24 preschool children with CLP and CPO. Subjects ranged in age from 34 to 63 months and were excluded from the study if they had evidence of mental retardation, a hearing loss of 30dB or greater, a bilingual home, were an adopted/foster child, attended nursery school, or received speech therapy. He also examined the non-linguistic factors of social economic status (SES), hearing, age at surgery and length of hospitalization, and their relationship to vocabulary measures. The PPVT was administered in a standardized fashion to obtain a measure of vocabulary comprehension, and was also modified to obtain a measure of vocabulary usage. Overall results indicated that all groups exhibited better comprehension than production; however children with cleft palate were slightly delayed in their development. While an attempt was made to control for syndromes associated with cognitive delay, no cleft type distinction was made, thus making it difficult to attribute the results of this cognitive delay (without medical genetic testing, many syndromes may have gone undetected).

Fox et al. (1978) examined the language and overall development by administering the Denver Developmental Screening Scale (DDS) (Frankenburg & Dodds, 1969), the REEL (Bzoch & League, 1971) and the Birth-3 Scale (B-3S) (Bangs & Garrett, 1973) to 24 children with CLP and CPO and a control group. Subjects ranged in age from 2 to 33 months and no effort was made to control for variables such as syndromes associated with cognitive delay, English as a second language, age of primary palatal surgery, etc. The authors considered the following factors which previous research had been found to affect the development of children with cleft palate: severity of the cleft, presence of middle ear problems, length of hospitalization and body weight at the time of testing. Among these variables, severity of the cleft was the only variable to reach statistical significance.

Results indicated that children with cleft palate scored 1-3 months below their non-cleft counterparts on all measures. The expressive language scores on the REEL were found to best differentiate the cleft palate group from the control group. Given that there were no controls for syndromes associated with cognitive delay and that no cleft type distinction was made, it is difficult to attribute the results of this study to the factors associated with clefting alone.

Some studies examined the environmental factors such as SES, length of hospitalization, etc. (Broen et al., 1998; Nation, 1970; Fox et al. 1978; Jocelyn et al. 1996) while others failed to address these factors (Smith & McWilliams, 1968; Dalston, 1990; Richman, 1980; Richman & Eliason, 1984). It is possible that the conflicting results may be attributed to the lack of control of these confounding variables. Longitudinal research in the area of normal language devlopment has attempted to identify risk factors which predict poor communication outcomes. The following are risk factors associated with poor communication outcomes: mother's education (Tomblin, Smith and Zhang, 1997), birth order (Tomblin, Hardy & Hein, 1991), parental history of speech and language (Tomblin et al, 1991), and low birthweight (Aram, Hack, Hawkins Weissman & Borawski 1991). Nelson (1998) provides a summary of the literature in terms of prognostic factors associated with better and poorer outcomes. In addition to the factors mentioned above, Nelson cites studies which report poorer prognosis for children: with a high-risk birth, who are older when first identified, who are older when still having difficulty, and who have problems with language or mixed articulation and language. She also notes that "when language disorder is mixed with other impairments (i.e. hearing loss, mental retardation)...the prognostic picture becomes even more complicated" (pg. 210).

An additional source of variability found in the cleft palate studies could have resulted from the lack of control for syndromes. In addition, many studies failed to make a cleft type distinction, thus making it difficult to determine whether children with CPO and

undetected syndromes were contributing to the overall report of delay in language acquisition skills.

Reading Abilities

Richman, Eliason and Lindregen (1988) examined the reading abilities of 172 children with CPO and CLP who ranged in age from 6 to 13 years old. In an effort to gain a representative sample from a large treatment centre, no attempt was made to control for speech and hearing characteristics or syndromes associated with cognitive delay. Children were assessed using the Standard Reading Inventory (SRI) (McCracken, 1966) and measures of word recognition and comprehension abilities were calculated. The authors defined reading disability in terms of performance below one standard deviation (standard score of 85) on the SRI. Using this definition they determined that the overall prevalence rate of reading disabilities was 35%. The results indicated that as a group, younger children performed more poorly than older children on comprehension and word recognition tasks. In terms of cleft-type differences, children with CPO had poorer scores regardless of age. Further analysis examined the relationship between word recognition and comprehension scores. Overall the results indicated that 18% of the sample had higher word recognition scores than comprehension scores while 6% of the sample had higher comprehension scores than word recognition scores. The authors suggested that this last subgroup may have had peripheral speech production abilities which were contributing to difficulties in the phonetic decoding process. While this study provides much needed data on the prevalence of reading disabilities in children with cleft palate, it does not address the variables which may contribute to these difficulties. Confounding variables such as mother's education, family history of reading difficulties and the extent of otitis media and hearing loss may be contributing factors to reading disabilities in this

population. Of particular interest is the subgroup who experienced difficulties which were apparently associated with peripheral speech difficulties. Further research is needed to determine the nature of these difficulties. Are the difficulties related to speech processing abilities, i.e. metaphonology, memory, language? This study indicates that children with CPO had poor scores across the age range. Given the recent advances in medical genetics, it would interesting to examine these children to determine whether in fact they do have a previously undetected syndrome. This study raises several questions regarding the nature of reading disability in this population and further research is needed to determine whether factors other than cleft palate are contributing to these difficulties.

To date, only one study has examined the word recognition errors of children with cleft palate. Stackhouse (1982) compared the word recognition, spelling and non-word matching abilities in children with cleft palate to a group with developmental verbal dyspraxia. Subjects ranged in age from 6;8 to 11;4 and the groups consisted of 10 children with cleft lip and palate, 10 children diagnosed with dyspraxia and a control group of 20 children. The cleft palate children were selected on the basis that they were currently receiving speech therapy for resonance or articulation problems. No information was provided concerning the distribution of cleft type, extent and nature of hearing loss, or degree of residual structural problems. The study controlled for IQ, English as a primary language, and physical handicap. Quantitative results indicated no difference between the cleft palate group and control group on the mean scores of reading and spelling and nonsense word matching. Significant differences were found between the dyspraxic group and the control group on all three measures. A qualitative analysis of the errors made by the cleft palate group revealed that they still demonstrated an understanding of segmentation and grapheme-phoneme conversion rules. Their errors did not differ substantially from those of the control group. On the other hand, the errors made by the dyspraxic group were described as delayed and deviant. Stackhouse concludes that the differences between

the cleft palate and dyspraxic groups indicate that a purely phonetic disorder does not interfere with reading and spelling abilities. This study appears to be in conflict with Richman et al. (1988) study which indicated that 6% of children with cleft lip and palate had lower word recognition scores than comprehension scores. Further research is warranted given the limitations of this study (i.e. relatively small number of subjects in the cleft palate group, the lack of information regarding cleft type, and speech and hearing status). One could assume that the small subgroup in the Richman study had more phonologically based errors (vs. phonetically based errors) which were impacting on their speech processing abilities in the word recognition process. However if one assumes that phonological processing difficulties are a part of an overall language processing deficit, then one would expect comprehension to be affected. Additional information concerning the metaphonological and language abilities, and family history of speech and reading difficulties is needed to gain a better understanding of the reading abilities of children with cleft lip and palate.

With this background on the speech, language and hearing abilities of children with cleft palate, we now turn to a more specific body of literature that discusses the development of metaphonological abilities.

METAPHONOLOGY

In recent years there has been considerable interest in the metaphonological abilities of language normal and language or literacy disordered children. Metaphonological awareness refers to the ability to access and make explicit one's tacit understanding of the speech sound structure of language. It is comprised of different aspects of linguistic knowledge such as segmenting words into syllables, producing rhyming words, and manipulating phonemes etc.

Task Types

Over the years phonological awareness has been defined and measured using a wide range of procedures available from research studies and published tests. Several researchers have noted that tasks, which claim to be tapping phonological awareness, vary in terms of the level and type of processing required and the different levels of metalinguistic and linguistic knowledge (e.g. Stackhouse & Wells, 1997; Stahl & Murray, 1998). Some of the common types of tasks include: rhyming, alliteration, phoneme blending, phoneme segmentation, phoneme manipulation and auditory discrimination.

Stackhouse and Wells (1997) provide a pscyholinguistic framework to evaluate various tasks. They cite the importance of asking the following questions: Is this an input or output task, Does the child have the lexical representations for the stimuli used in the task? Does the child have to access these lexical representations in order to complete the task? They also provide the following cautions:

> Tests of seemingly different skills (e.g. rhyme and blending) can tap the same level of processing. Tests of seemingly the same skill (e.g. rhyme tests) can tap different levels of processing (e.g. depending on if pictures are involved, if a spoken response is expected). Tests can be analyzed in terms of (a) how they are presented (b) the test stimuli involved and (c) the response required. (pg. 73)

The wide range of tasks and procedures used to assess phonological awareness has contributed significantly to the variability found in the literature. The following section

provides a summary of the current literature on the development of phonological awareness skills.

Development of Phonological Awareness Skills

Like other speech and language skills, phonological awareness develops from emergence to mastery. Many studies have attempted to delineate what constitutes normal development of metaphonological skills. Unfortunately this research has yielded inconsistent findings. Differences in task variables and definitions of phonological awareness have contributed to the varied findings. What follows is a critical review of the literature to date.

Overall the available evidence suggests that children typically become aware of onset and rimes before they develop comparable sensitivity to phoneme units (e.g. Bowey and Francis, 1991; Bowey, 1994; Bernhardt, Edwards &Rempel, 1995). In her 1994 study Bowey found that preschool children who were beginning to read score higher on phonological sensitivity tasks than non-readers. This study involved two tasks (1) thinking about sound structure (phonological oddity tasks) and (2) thinking about phonemes (phonological identity tasks). To ease the phoneme identity tasks, pictures with labels were used. The results showed that preschool children who are beginning to read scored higher than non-readers on phonological sensitivity tasks. In non-readers, phonological sensitivity is associated with letter knowledge and verbal ability. Children who were entering reading instruction could not make judgments about phonemic overlap in spoken words, although many were sensitive to subsyllabic onset-rime structure.

There is evidence to suggest that orthographic onset and rime units serve as functional units of word recognition in skilled readers (Bowey, 1990, 1996; Coltheart & Leahy, 1992). However, some claim that onset and rime are merely units of working memory. Treiman and Chafetz (1987) provide the first unambiguous demonstration of orthographic rimes as functional units of word recognition. This study involved a lexical decision tasks with tokens like (1) CR//ISP (2) CRI//SP. Results indicated faster lexical

decisions to words which were segmented between the onset and rime units. This suggests that end chunks of words can be considered units of print and not just units of verbal working memory. Bowey (1990, 1996) also found evidence that adults use orthographic rime correspondences rather than grapheme-phoneme correspondences when these two conflict.

Chafouleas, Lewandowski, Smith and Blachman (1997) attempted to determine the developmental progression of phonological skills. In their methodological review of early studies, they note the following limitations: (1) phonological awareness was measured using one or two tasks over large age groups, or (2) phonological awareness was measured using several tasks with one age group. The researchers were motivated by the lack of data on multiple tasks across multiple ages. Their subjects included typically developing children in kindergarten, grade one and grade two. Using various tasks they concluded that phonological awareness develops in the following order: rhyme, alliteration, phoneme blending, phoneme segmentation, and phoneme manipulation.

Wagner, Torgeson and Rashotte (1994) also examined multiple tasks across multiple ages and they found a similar progression: sound isolation, sound categorizations, blending (real words), deletions, and segmentation.

Factors Affecting Phonological Awareness Ability

There is evidence which suggests that performance on phonological awareness tasks can be influenced by several factors. This section will examine the relationship between phonological awareness and the following factors: effect of previous literacy experience, memory, speech perception, cognitive ability, and language abilities (phonology and morpho-syntax).

Effects of Literacy Experience on Phonological awareness

Considerable attention has been focused on the question: Does the ability to manipulate speech sounds depend on knowing alphabetic writing?

The following studies provide evidence that sensitivity to oral structure is affected by alphabetic literacy. Morais, Cary, Algeria and Bertelson (1979) tested illiterate and late literate Portuguese adults on their ability to manipulate sounds. The subjects were assigned to one of two metalinguistic tasks; phoneme deletion, and phoneme addition. Each task involved words and non-words. The late literates found both tasks much easier to do than the illiterates. The evidence suggests that one's ability to manipulate sound units of speech is restricted if one has never learned to read. Read et al. (1986) later predicted that the inability to manipulate sounds was due to an inability to read an alphabetic script. Read tested Chinese readers who were alphabetically illiterate and alphabetically literate, finding that the alphabetically literate subjects performed better than the illiterate subjects. The results of this study suggest that learning to read an alphabetic script system makes us able to think of sounds at the phoneme level.

The ability of the illiterate subjects to manipulate multi-phonemic units as opposed to single phonemes is congruent with the findings in preschool children. Evidence from young children suggests that the ability to make judgments about sound patterns in spoken words is partly a product of learning to read an alphabetic script. If the ability to recognize phonemes is something you develop during the experience of learning to read, then there should be no difference in older and younger students in grade one. Age does not make a difference, but schooling does. To test this hypothesis Bowey and Francis (1994) studied older preschool kids, young grade ones and older grade ones. The participants were given three phonological oddity tasks (rime, onset and phoneme). The older preschool children performed less on these tasks. It appears that going to school increases the awareness of sound structures in general.

These adult and child studies support the hypothesis that reading and phonological awareness have a reciprocal relationship. The next section examines the role of cognition in relation to phonological awareness abilities.

Cognitive Abilities

While there is evidence to suggest that deficits in verbal working memory and speech perception contribute to the variance in performance on metaphonological awareness tasks, questions remain regarding the relationship between cognitive ability and metaphonological awareness.

There has been little research investigating the specific cognitive skills which are prerequisites for developing metaphonological skills. The majority of studies have explored the relationship between IQ scores and performance on metaphonological awareness tasks. For example, McBride-Chang (1996) used the Wechsler Intelligence Scale for Children (WISC) (Wechsler, 1974) as a measure of general cognitive ability. In this study she examined the relationship between speech perception, phonological awareness and cognitive ability. The results of the multifactor analysis revealed that general cognitive ability was a strong predictor of phonological awareness. However, Magnusson and Naucler's (1990) investigation of language disordered and language normal children, found that cognitive abilities only played a role in terms of the language disordered children's ability to perform on segmentation tasks.

These studies demonstrate that there is disagreement about the role of general cognitive ability in phonological awareness. Considerable attention has been paid to one aspect of cognitive functioning: working memory. The following section examines the relationship between phonological working memory and phonological awareness ability.

Memory

Some research has pointed to phonological working memory as a factor which is related to performance on metaphonological awareness tasks (Gillam and VanKleeck, 1996; Webster, Plante & Couvillion, 1997; Gathercole & Baddeley, 1993). The concept of phonological working memory is based on a model of working memory outlined by Gathercole and Badderly (1993). This model incorporates modality-specific storage components: the phonological loop, the visual-spatial sketch pad and the central executor. The phonological loop is the auditory storage mechanism, while the visual-spatial sketch pad is the visual storage mechanism. The central executor can be used either for storage or processing and is considered the main component because it regulates and co-ordinates information. The phonological loop has been described as consisting of two components: phonological coding and phonological recoding. Gillam and Van Kleeck (1996) suggest that the phonological coding component involves short-term storage of phonological representations which decay with time. In order to prevent decay, the recoding component serves to maintain the phonological representations through a rehearsal process. Performance on metaphonological tasks have been explained within the framework of this model. For example, in an identification task used in the Test of Phonological Awareness (Torgeson & Bryant, 1994) (i.e., jeep, dog, pig, bag, which word has a different last sound?) the first step involves activating the phonological representation for each of the words. These representations must be kept "alive" in the short term storage while the listener processes the sequence of phonemes in order to respond to the question.

Webster et al.(1997) claim that the phonological deficit hypothesis can account for the poor phonological awareness skills of children with phonological impairment. The authors suggest that these children may have weak or incomplete phonological representations, inefficient rehearsal of verbal information, reduced memory capacity or difficulties accessing phonological representations. In turn these deficits in the phonological system impair phonological processing abilities. Similar theories have been

posited for children with language disorders. Gathercole and Baddeley (1993) suggested that children with language disorders may have rapidly decaying phonological traces which make the storage of phonological information difficult. The most commonly used tests of verbal working memory are digit span and word span tests.

The next section further examines the notion of phonological representation as it concerns speech perception skills.

Speech Perception

Speech perception abilities have been named as an additional factor which may account for differences in performance on phonological awareness tasks. Manis, McBride-Chang, Seidenberg, Keating, Doi, Munson and Petersen (1997) found that children whose phonemic boundaries are robust performed better on phonological awareness tasks. They limited their study to the a phoneme identification paradigm in an attempt to reduce memory demands. These results provide support for the phonological deficit hypothesis because they indicate that deficits in speech perception abilities interfere with the formation of robust phonological representations.

McBride-Chang (1996) also used an identification task to study the relationship between speech perception and phonological awareness abilities. In this task only one stimulus at a time was considered, and a forced choice paradigm was used to avoid confounds in measurements such as articulation ability. Voice onset time was manipulated in the first two tasks as children were required to identify when the targets bath/ path or slit/split were spoken using synthesized speech. The third task involved identifying when the manner of articulation changed in the non-words ba/wa. The results of a factor analysis revealed that once verbal short term memory and naming abilities were controlled for, speech perception accounted for a considerable amount of the variance on the phonological awareness tasks.

These studies provide evidence that underlying speech perception difficulties may be contributing to phonological processing difficulties. The relationship between language ability and phonological awareness ability will now be examined.

Language Abilities: Morphology and Syntax

There is controversy in the literature surrounding the extent to which language skills influence performance on phonological awareness tasks. Unfortunately these studies are confounded by variables such as subject selection (some are chosen because they have phonological impairment whereas some are chosen because they have language impairment) and task selection (different tests measure different aspects of syntax and morphology). None of these studies have been replicated.

In addition to examining the phonological awareness abilities of 31 boys with expressive phonological impairments, Bird, Bishop and Freeman (1995) administered two language tests. The Action Picture Test (APT; Renfrew, 1980) was used to measure productive abilities, while the British Picture Vocabulary Scale (BPVP; Dunn, Dunn, Whetton & Pintilie, 1982) measured comprehension abilities. Based on their performance on the language tests, subjects were grouped according to the existence of a concomitant language disorder. The authors suggest that: "If a general linguistic impairment were responsible for depressing performance on the phonological awareness task, we would expect to see few problems in those children with a relatively pure speech disorder" (pg. 459). The results indicated that both groups had difficulties with phonological awareness tasks and the authors reject the claim that syntactic and vocabulary skills influence performance on phonological awareness tasks.

These findings are not consistent with those found by Magnusson and Naucler (1990). This longitudinal study explored the relationship between metaphonological abilities, language abilities, short term memory, reading and spelling. Subjects included 74 Swedish children selected on the basis of a previously diagnosed language disorder, and

37 children with normal language development. Data were collected before children were given formal reading instruction, and four more times until the end of fourth grade. Results indicated that two metaphonology tasks (phoneme identification and rhyming) were correlated with language competence. A closer inspection of the data indicated that not all children with language disorders experienced difficulties on phonological awareness tasks. Unfortunately a detailed description of the tasks involved in the language comprehension and syntactic production tests administered is not provided. Therefore, it is not possible to comment on the nature of language deficits involved in phonological awareness.

Major and Bernhardt (1998) used the Index of Productive Syntax (IPSyn) to measure morphosyntactic skills in 19 preschool children with phonological delay. They found a moderate relationship between performance on phonological awareness tasks and morphosyntactic production.

Chafouleas et al. (1997) included measures of general language abilities in their study examining the development of phonological awareness abilities. The language measures included the Peabody Picture Vocabulary Test - Revised (Dunn & Dunn, 1981), a letter-word identification test and letter names and letter-sound tasks. The results indicated that vocabulary comprehension was moderately related to performance on phonological awareness tasks. It is of interest to note that subject selection for this study was not based on previous history of speech or language delay because the main goal was to determine the developmental progression of phonological awareness skills.

Kahmi, Lee and Nelson (1985) examined the metalinguistic abilities of children with and without language disorders. Subjects included 45 children aged 3 to 6 years old, and were selected based on previous history of language delay. Control groups were matched for mental age and language-age. Three measures of metalinguistic awareness were administered: dividing sentences into words, dividing words into syllables and dividing words into phonemes. Overall, the results indicated that children with language

disorders performed more poorly on all three tasks than their mental-age and language-age matched counterparts.

A recent study by Nathan, Stackhouse and Goulandris (1998) was an attempt to identify whether children with specific phonological problems and accompanying language problems had more difficulties with phonological processing tasks than those with speech-only problems. Subjects included 47 4-5 year olds and were selected on the basis of existing speech difficulties as determined by scoring one standard deviation below the means on a standardized articulation test. All had a nonverbal IQ within normal limits. A control group was matched based on chronological age, gender and non-verbal IQ. The following tests were administered to measure language comprehension and production and speech processing abilities: Test for the Reception of Grammar (Bishop, 1983), British Picture Vocabulary Scale (Dunn et al. 1982), Renfrew Action Picture Test (Renfrew, 1988), The Renfrew Bus Story (Renfrew, 1995), Naming Test (Snowling, 1998), word and non-word repetition and an auditory-lexical task. Groups were divided in terms of presence versus absence of a language problem. While this study did not attempt to measure phonological awareness, the results indicated that children with both speech and language problems performed worse than those with speech only problems on measures of speech processing abilities. The authors suggest that children with specific speech output difficulties do not necessarily have input deficits affecting their phonological representations. However, if language problems co-occur, then input processing problems are likely to be present.

There appears to be an overall tendency for language delay to influence metaphonological ability; however it is not clear as to which language skills are related. More research is needed to find out which language skills contribute to the variance in metaphonological abilities. The next section will examine the relationship between phonological ability and phonological awareness.

Language Abilities: Phonology

The connection between phonological awareness abilities and phonological impairment has been explored in a few studies. Webster and Plante (1992) used the phonological deficit hypothesis to predict that children with phonological impairment are at risk for deficits in phonological awareness. They hypothesized that these children would have difficulties inputting information into working memory because the phonological impairment interferes with the subvocal speech rehearsal process. The study involved 22 children aged 6;5 to 8;6. Subjects were selected on the bases of normal phonological development or moderate to severe phonological impairment and all had normal nonverbal intelligence. The following tests were administered to assess language comprehension and production, word recognition and metalinguistic awareness: Peabody Picture Vocabulary Test-Revised (Dunn & Dunn, 1981), the Test of Auditory Comprehension of Language-Revised (Carrow-Woolfolk, 1985), the Word Recognition test from the Stanford Achievement Test (1972), and the Test of Awareness of Language Segments (TALS) (Sawyer, 1987). The group with phonological impairment performed significantly worse than the control group on the TALS. (This test is a measure of metalinguistic awareness because it involves segmenting sentences into words, segmenting words into syllables and segmenting words into phonemes.) Despite the limited sample size, the results provide preliminary support for the phonological deficit hypothesis because children with phonological impairment experienced difficulties with phonological awareness tasks.

In a later study, Webster et al. (1997) sought to further test the phonological deficit hypothesis by increasing the sample size, and measuring verbal working memory. This longitudinal study examined the relationship between phonological impairment and literacy outcomes. Baseline measurements of phonological awareness, working memory and prereading ability were taken and used to evaluate progress. An additional attempt was made to examine the relationship between productive syntax-morphology and phonological awareness. Subjects included children, 29 of whom were classified as phonologically

impaired while 16 had normal phonology. Fifteen of the children with phonological impairment were matched with normals for mental age and gender. Phonological impairment was operationally defined using the percentage of consonants correct (PCC) procedure. In addition the Khan-Lewis Phonological Analysis test (Khan & Lewis, 1986)was used to generate a severity rating. The results of group mean comparisons indicated that children with phonological impairments performed worse than their normal peers on the following tasks: sentence memory, pseudoword segmentation and letter identification. A regression path analysis revealed that sentence memory alone was the best predictor of letter identification: it explained 28% of the variance. In addition they found that there was a strong relationship between productive phonology and verbal working memory. The authors conclude that " overt developmental phonologic impairment (reflected in speech intelligibility levels) may hinder the establishment of secure, stable phonological representations" (pg. 373). The authors suggest that the results support the phonological deficit hypothesis.

In their longitudinal study, Bird et al. (1995) found further evidence that children with phonological impairment have difficulties with phonological awareness tasks. Subjects included 31 boys aged 5;0 -7:4 who were being treated for phonological impairment. A control group included 15 children with normal phonology who were matched for nonverbal ability and age. Phonological impairment was assessed using a nonstandard measure which used pictures to elicit single words. The test was transcribed and scored using the percent consonant correct method (Shriberg & Kwiatkowski, 1982). Results indicated that 23% had mild-moderate disorder, while 29% had a moderate-severe disorder and 48% had a severe disorder. Additional tests were administered at T1 to assess language comprehension and production, and phonological awareness. A number of literacy outcome measures were administered at T2 and T3. The authors chose to measure phonological awareness using the following non-verbal tasks: rime matching, onset matching, and onset segmentation and matching. Results indicated that compared to their

normally developing counterparts, the children with phonological impairment had difficulties with the phonological awareness tasks and had later problems with reading. The authors hypothesis that poor performance on the phonological awareness tasks is a result of the children with phonological impairment failing to analyze syllables with respect to their phonemes. They suggest that the children analyze syllables as whole chunks. The results lend further support to the theory that children with phonological impairment have incomplete phonological representations of words (or difficulties accessing the representations) that hinder phonological processing abilities.

In a study examining effects of phonological and metaphonological intervention Bernhardt and Major (1998) examined the phonological awareness abilities of 19 preschool children with moderate to severe phonological disorders. Phonological ability was evaluated using the following aspects of non-linear phonology: segmental, syllable and word shape profiles. Before intervention, results indicated a relationship between phonological development and performance on metaphonological tasks. Individual variation was noted and the authors suggested that "children's ability to perform well on tasks seemed to be related to several factors, including their age, and the level of severity of both their morphosyntactic and phonological disorders" (pg. 440).

In a recent study by Larrivee and Catts (1999) which examined the early reading skills of kindergarten children with expressive disorders, the authors suggested that the severity of the phonological disorder was a factor. The results indicated that children who had more severe phonological disorders performed significantly worse on phonological awareness and language measures and had poorer reading outcomes. Expressive Phonology was measured using the Arizona Articulation Proficiency Scale (AAPS) and a task which involved the repetition of multisyllabic words and nonwords. Both measures were transcribed and scored using the percentage consonant correct method. The authors noted differences in results based on the phonology measure used. The multisyllabic task proved to account for more variance in the reading outcomes measures than did the AAPS.

The authors raise concerns regarding the potential bias of subject selection. In this study subjects were selected on the basis of expressive phonology and they found only a slight relationship between poor outcomes and poor language ability. On the other hand studies which select on the basis of language impairments find a stronger relationship.

These studies suggest that severity of the phonological disorder and morphosyntactic ability place a role in determining the relationship between phonological awareness and language abilities. In addition, there is evidence to suggest that cognitive factors such as verbal working memory and speech perception skills, and prior literacy experience affect performance on phonological awareness tasks.

The Present Study

The present study was motivated for several reasons. As previously mentioned, the original reason was in response to Richman et al.'s (1988) work which reported a higher than average prevalence rate of reading disorders among children with cleft palate. Given the link between phonological awareness and later reading difficulties, there was a need to document the metaphonological abilities of this population. Information regarding phonological awareness performance and verbal working memory will contribute to the literature on phonological processing abilities in this population.

The Richman et al.(1988) study raised further questions regarding cleft-type differences. For the most part, the body of literature outlining language development in children with cleft palate fails to report cleft-type differences. In addition subject selection methods have not always excluded children who have syndromes associated with cognitive delay. Given the recent advances in medical genetics, it is now possible to detect more of these syndromes than was previously possible (Shprintzen & Goldberg, 1995). Therefore this study was motivated to improve the subject selection process and to report the language abilities of children without known syndromes.

In addition, previous studies have failed to control for environmental factors (such as mother's education and family history of speech and language problems) which may be contributing to the language delay in this population. Given the claims that phonological impairment may hinder the establishment of secure and stable phonological representations (Webster & Plante, 1997) and the prevalence of phonological disorders within the cleft palate population, there is a need to document the nature of the speech disorder. To determine the nature of the speech disorder one must examine the types of errors produced, i.e., do they reflect deficits in the structural integrity of the oral mechanism or do they reflect phonological problems?

The questions regarding the nature of the metaphonology, language and phonology abilities motivated the following questions and their associated hypothesis:

1. Do children with a history of cleft lip and palate perform below normal limits on standardized tests of morphosyntax and vocabulary, phonology and metaphonology?

Null Hypothesis: Children with cleft palate will perform within normal limits on standardized tests of language, phonology and metaphonology.

2. Qualitative Analysis: What is the nature of speech disorders within this particular group of children with cleft palate?

3. Is there an interaction between environmental factors, IQ, structural integrity, metaphonology, phonology, and performance on language tasks?

Null Hypothesis: There will be no relationship between environmental factors, IQ, structural integrity, metaphonology, phonology and language measures.

4. Is there an interaction between environmental factors, IQ, structural integrity, language, phonology, and performance on metaphonology tasks?

Null Hypothesis: There will be no relationship between environmental factors, IQ, structural integrity, language, phonology, and metaphonology measures.

In response to the body of literature which suggests that earlier palatal repair equals better speech outcomes the craniofacial clinic at BCCH has adopted the practice of

providing early primary palatal surgery. In light of this decision to provide early primary surgery, this study was also motivated by the need to examine the speech production abilities in relation to the age of primary palatal repair.

5. Does early palatal repair influence speech, language and metaphonological outcomes?

Null Hypothesis: The correlation between early palatal repair and speech and language outcomes is zero.

6. Do residual structural problems influence speech, language and metaphonological outcomes?

Null Hypothesis: The correlation between structural problems and speech and language outcomes is zero.

Given that children with cleft palate may be more at risk than their cleft palate only counterparts for having an undetected syndrome associated with cognitive delay, an attempt was made to report cleft-type differences.

7. Do children with cleft palate only perform worse on standardized measures than children with cleft lip and palate?

Null Hypothesis: There will be no difference in language test performance in children with cleft palate only and cleft lip and palate.

CHAPTER TWO: METHOD

SUBJECTS

Twenty subjects participated in this study; six subjects (all female) with Cleft Palate Only (CPO) and fourteen children (6 females and 8 males) with cleft lip and palate (CLP). All subjects are followed by a cleft-palate team at a local children's hospital. A pool of 156 potential subjects (Kindergarten-grade one age) was identified by a review of the hospital's cleft palate database. From that pool, 103 subjects were eliminated. The criteria for exclusion was (1) an identifiable syndrome associated with cognitive delays (2) sensorineural hearing loss (3) those whose primary language is not English (4) weight less then 5 pounds at birth and premature or 5 minute Apgar score of 6 or less (5) distance from university. Letters of recruitment were forwarded to 53 families. Twenty families consented to participate in the study. Table 1 describes the clinical characteristics of the sample.

Table 1. Clinical Characteristics

	Mean	SD	Minimum	Maximum
Age (months)	74.6	6.5	66	84
Birth weight (g)	3493	426.8	3007	4200
Apgar (1 minute)[a]	8.5	.7423	7	10
Apgar (5 minutes)	9.2	.4571	9	10

[a] Apgar scores were available for 15/20 subjects

Information on a range of background variables was collected; including current grade, birth order, maternal education (see table 2) family history of speech and language impairment and reading difficulties (see table 3). Medical records were reviewed and the following information noted: age of primary repair, age of secondary repair (when warranted), age and frequency of myringotomy tubes (see table 4), nature and degree of hearing loss, and history of speech and language therapy.

Table 2. Background Variables

	Number (/20)	Percent (%)
Current Grade		
Kindergarten (end)	12	60
One (end)	8	40
SES		
1&2[a]	14	70
3,4 &5	6	30
Maternal Education		
High school only	10	50
Vocational school or some college	4	20
Bachelor's degree	6	30
Birth Order		
First born	10	50

[a] 1 & 2 = highest (professional and semi-professional)

Table 3. Family History

	Number (/20)
Family history of speech and language difficulties	3 (S# 7,9,14)
Family history of reading difficulties	4 (S#2,7,9,17)

Table 4. Mean Age of Primary Palatal Repair and First BMT Insertion

	Mean	SD	Minimum	Maximum
Age of first BMT[a] insertion (months)	10.58	1.80	9	14
Age of Palatal repair (months)	10.10	1.33	7	12

[a] Bilateral myringotomy tubes

The mean age of palatal repair (10 months) reflects the local clinic's approach of early palatal repair. In addition, the clinic has adopted the practice of inserting ventilation tubes at the same time as the initial palatal repair. Timing of ventilation tubes placement was examined relative to the timing of primary palatal surgery; 90% had ventilation tubes placed before or at 12 months old. The mean number of times tubes were placed was 2.3 and ranged from 0 (S#2)to 5(S#14). At the time of primary palatal surgery 18 of the 20 (90 %) had ventilation tubes placed. (S#14 had tubes inserted 1 month following primary palatal repair due to administrative problems, while S#2's parents did not consent to BMT placement).

Cognitive Ability

Subjects had an average non-verbal cognitive score as determined by the Test of Non-Verbal Intelligence (TONI-3) (Brown, Sherbenou & Johnsen, 1997) or the TONI-2 (Brown, Sherbenou & Johnsen, 1990) for the 5 year olds. The TONI-2, and TONI-3 are non-verbal tests requiring perceptual judgment. The subjects had a mean score of 106 (SD=10.67) scores thus ensuring that their functioning was specific to cleft palate and not

just the result of a low IQ. See Appendix A for a more complete description of this test, and table 5 for mean scores.

Memory

Each subject had an average verbal working memory score as determined by the Wechsler Intelligence Scale for Children-III, Digit Span subtest (Wechsler, 1991). The subjects had a mean score of 9 (SD=2.4), thus ensuring that their ability to perform on language and metaphonological tasks was not the result of poor verbal working memory. The digit forward and digit backward scores were combined to obtain a digit span total. See Appendix A for a description of this test, and table 5 for mean scores.

Table 5. Mean Scores for Memory and Cognitive Measures

	Mean	SD	Minimum	Maximum
Digit span (WISC raw score)[a]	9	2.427	6	14
TONI (Quotient)[b]	106	10.67	93	136

[a] Standard scores were only available for the 6 year olds (N=13, mean 9.46, SD 2.817, min. 7, max. 15).

[b] TONI-2 scores are reported for the 5-year-old subjects, and TONI-3 scores for the 6-year-olds.

TESTING MATERIALS

Standardized Tests

Subjects were given six standardized tests in order to assess speech and language abilities, metaphonological ability, verbal working memory ability, and cognitive ability.

The Photo Articulation Test - 3 (Lippke, Dickey, Selmar and Soder, 1997) was used because it provided normative data by age and gender. The word list for this test included a variety of segments and wordshapes. Vocabulary and morphosyntactic abilities were assessed using the Test of Language Development - P:3 (Newcommer and Hamill, 1997). This commonly used tests was administered because previous studies have found subtests (i.e., sentence imitation) to be predictive of future reading difficulties (Catts, Fey, Zhang & Tomblin,2000). Two tests were used to gain a profile of children's phonological awareness abilities. The Test of Phonological Awareness (TOPA) (Torgeson et al., 1994) was used to measure the awareness of individual sounds in words. TOPA scores have been found to be related to reading growth in the first grade. The Phonological Awareness Test (Robertson & Salter, 1995) was used as it provided normative data and consisted of a variety of tasks which have been related to later literacy skills (i.e. letter identification, see Catts et al. 2000). The following subtests were administered: Rhyming Discrimination and Production, Blending: Syllables and Phonemes and Letter Identification. The digit span subests of the Wechsler Intelligence Scale for Children III (Wechsler, 1991) was administered because previous research has used this as a measure of verbal working memory (Gathercole & Badderly, 1993; Webster et al. 1997). The Test of Nonverbal Intelligence-2 (Brown, Sherbenou & Johnsen, 1990), and the Test of Nonverbal Intelligence-3 (Brown, Sherbenou & Johnsen, 1997) were used to test intelligence, aptitude and reasoning skills without verbal instruction See Appendix A for a description of these tests.

Hearing Screening

Hearing was screened using a calibrated portable audiometer Maico MA 40. The screening procedure was based on the procedure outlined by the Vancouver/Richmond Health Department (See Appendix B). Hearing was screened at 20 dB across the following frequencies: 500, 1000, 2000, 4000 Hz, bilaterally. Given the history of

recurrent otitis media with this population, the screening was administered to ensure that the subjects had adequate hearing levels for the purposes of speech and language testing. All subjects passed the hearing screening.

Oral Mechanism Exam

The oral structure of each subject was appraised using a modification of the Dworkin-Culatta Oral Mechanism Examination (D-COME, 1980). Observation of the following structures and functions were noted:

1. Presence of Fistula: yes, no, location

2. Occlusion Data: Class I, II or III

3. Teeth: Vertical relationships: normal, open bite, close-bite, cross-bite, end-to-end bite

   Cutting edges: normal, rotated, jumbled, missing, extra

   Gaps: yes, no

4. Contour of Palate: Normal, flat, deep and narrow

5. Tongue Function: elevation

   deviation to left and right

   presence of tongue tie? yes, no

The examiner made clinical observations which were later confirmed by consulting with the most recent orthodontic and speech pathology reports.

TESTING

Individual testing took place over two 90-minute sessions with a family member present; with the exception of three out-of-town subjects whose sessions lasted 2 1/2 hours. Tests were administered in one of three assigned orders to eliminate the test-order effect and in standardized fashion according to the procedures outlined in the various test manuals. Upon completion of the session, the caregiver was asked to fill in a questionnaire in which they reported family history of speech and language difficulties, birth-order, mother's education, socio economic status etc. (See Appendix C.)

All sessions were audiotaped for the purposes of transcription and reliability checks. A Marantz tape recorder with a Realistic PZM (table top) microphone was used to record performance.

Table 6 outlines the average standard score for the given tests.

Table 6. Scoring Criteria

TEST NAME	# of ITEMS	AVERAGE STANDARD SCORE
TOLD-P:3 (1997)		
Subtests	20-30 [a]	10 +/- 3
Quotient		100 +/- 15
TONI-2,3 (1990, 1997)	45	
Quotient		100 +/- 15
TOPA (1994)	20	
Standard Score		100 +/- 15
Phonological Awareness Test (1995)		
Subtests	10	100 +/- 15
Mean of Standard Scores (TOPA + Phonological Awareness Test)	60[b]	100 +/-15
PAT-3 (1997)	70	100+/-15

[a] The number of items per subtest is as follows: Picture Vocabulary-30, Relational Vocabulary-30, Oral Vocabulary-28, Grammatic Understanding-25, Sentence Imitation-30, Grammatic Completion-28, Word Discrimination-20.

[b] The raw score for the Metaphonology Measure was calculated by adding the raw scores of each individual subtest, with the exception of the TOPA and the letter identification subtest. The raw scores of 20 in these tests were divided by 2 thus creating a total possible raw score of 60.

The phonological sample was narrowly transcribed and scored according to the procedures outlined in the PAT-3 manual, in addition it was scored in accordance with Trost's (1981) error classification system.

The oral mechanism exam was scored by in the following manner: each category was judged to be abnormal or normal based on at least one abnormal sign per category. A

normal category was given one point, while an abnormal category was given two points. The following categories were included: presence of fistuale, occlusion, teeth, contour of palate, tongue function. A measure of overall structural integrity was constructed by combining the oral mechanism score with two additional variables: resonance and voice. The resonance variable was included and judged to be abnormal if speech was hypernasal, hyponasal (including cul-de-sac resonance) and or audible nasal air emissions were present. Abnormal resonance may be a reflection of a faulty velopharyngeal closure which can occur with or without structural deficiencies. The voice variable was included because some children with cleft palate experience laryngeal symptoms such as hoarseness, low speaking volume, strain or strangled voice quality.

SUMMARY

All twenty children, ranging in age from 66 months to 84 months, were administered standardized tests of language, metaphonology, phonology, non-verbal intelligence and verbal working memory. Background variables for each of the children and age of primary palatal repair and variables associated with hearing status were collected from medical records. All children passed the hearing screening and were within the average range for the verbal working memory and cognitive measures.

The following chapter looks at the performance on the language, metaphonology and phonology measures and at the relationships between the variables.

CHAPTER THREE: RESULTS

The primary goal of this research was to examine the speech, language, and metaphonological abilities of kindergarten and grade one children with cleft palate. This was done using standardized tests. The psychometric characteristics of the speech and language scores (e.g., skew and kurtosis, standard deviation ratios, correlations between means and standard deviations, small and or unbalanced cell sizes) demand analysis using non-parametric inferential statistics (Spearman rank correlations) when measures were treated as continuous variables; parametric statistics (means, standard deviations) were used for descriptive purposes. Correlation analyses were completed with both parametric and nonparametric statistics, reporting only the parametric findings when the nonparametric coefficients were essentially similar. An alpha level of .05 (two tailed) was used for all statistical tests. The following chapter will address each research question beginning with a comparison of children with cleft palate to standardized norms, multiple regression, surgery age,residual structural problems, and cleft type group distinction,

Language: Metaphonology, Morpho-syntax, Vocabulary and Phonology

Question 1: Do children with a history of cleft lip and palate perform below normal limits on standardized tests of language, phonology and metaphonology?
Null Hypothesis: Children with cleft palate will perform within normal limits on standardized tests of language, phonology and metaphonology.

A descriptive statistical analysis of the overall standard scores was done to answer the question of whether children with cleft palate performed within normal limits on standardized speech and language tests.

TABLE 7. Overall Standard Scores for the Language, Metaphonology and Phonology Measures

TEST	Mean	SD	Minimum	Maximum
TOLD P:3 (1997)	111.8	10.83	98.00	137.00
Metaphonology [a]	100.1	7.89	80.17	113.17
PAT-3 (1997)	76.4	19.17	60.00	113.00

a A composite score (mean standard score) for metaphonology was calculated by using six standard scores of the TOPA (1994) and The Phonological Awareness Test (1995) (each individual subtest score was used).

As this table indicates, the only score which deviates one standard deviation from the mean of 100 +/-15, is the PAT-3 (1997). A descriptive analysis of the articulation skills will be discussed later.

The TOLD: P-3 (1997) quotient score was used because there was no statistically significant difference between the Listening Quotient (standard scores from Picture Vocabulary and Grammatic Understanding subtests) and the Speaking Quotient (standard scores from the Oral Vocabulary and Grammatic Completion subtests). While it was observed that the majority of subjects fell within the average range of ability (mean=111.8, SD=10.83) inspection of individual subtests was warranted. Table 8 provides standards scores for the six core TOLD subtests and one supplemental test.

Table 8. Descriptive Statistics for Individual TOLD: P-3 Subtests

SUBTEST (scaled scores)	Mean	SD	Minimum	Maximum
Picture Vocabulary	12.2	2.69	8.00	17.00
Relational Vocabulary	11.65	2.18	8.00	15.00
Oral Vocabulary	11.25	1.58	9.00	16.00
Grammatic Understanding	11.4	2.34	6.00	15.00
Sentence Imitation	12.15	1.98	9.00	16.00
Grammatic completion	11.70	2.38	7.00	17.00
Word Discrimination	11.40	2.48	6.00	15.00

Inspection of the individual profiles indicates that overall, children with cleft palate performed within normal limits (10 +/-3) on the individual subtests. Not all individual subjects performed within normal limits on each subtest. A closer look at individual data indicates that one subject (S# 5) scored poorly (standard score of 6) on the grammatic understanding subtest, while another child (S# 17) scored poorly (standard score of 6) on the word discrimination subtest. See table 10 for individual data.

Table 9 provides standard scores for the six metaphonology subtests

TABLE 9. Descriptive Statistics for Metaphonology Tests

TEST (scaled scores)	Mean	SD	Minimum	Maximum
TOPA	97.65	12.31	76.00	116.00
Rhyming: Discrimination	102.8	11.68	73.00	113.00
Rhyming: Production	109.5	3.55	101.00	116.00
Blending: Syllables	91.85	11.70	61.00	112.00
Blending: Phoneme	97.20	14.20	68.00	123.00
Letter Identification	101.30	11.97	63.00	120.00

Table 10. Individual Profiles

Subjects [a]	Grammatic Understanding [b]	Sentence Imitation	Grammatic Completion	Word Discrimination	TOPA [c]	Rhyming: Discrimination	Blending: Syllable	Blending: Phoneme	Letter Identification	Digit Span	Phonology	Structure [d]	Family History [e]	Mother's education	SES [f]	Total
S 2													♦			1
S 5	♦									♦	♦			♦	♦	5
S 6				♦		♦					♦			♦	♦	5
S 7					♦		♦	♦					♦			4
S 8											♦	♦				2
S 9				♦	♦	♦	♦				♦		♦	♦	♦	8
S 10												♦		♦		2
S 11														♦		1
S 12			♦											♦	♦	3
S 14											♦	♦				3
S 15							♦					♦		♦	♦	4
S 17				♦	♦		♦	♦	♦		♦		♦	♦	♦	9
S 18						♦					♦					2
S 19										♦	♦			♦		4

[a] Results for S# 1,3,4,13,16 & 20 were all within normal limits
[b] Results for the TOLD: P-3 are recorded if were below average (i.e., standard score of 7 or less)
[c] Results for the TOPA, and the Phonological Awareness Test, PAT-3 and digit span are recorded if they were one standard deviation below the mean (i.e., standard score of 85 or less).
[d] Results for the Structure variable are recorded if there were more than three abnormal signs on the Oral Mechanism Exam.
[e] A positive family history of speech and language difficulties is reported and Mother's education is noted if grade 12 or below.
[f] Socio-Economic Status is noted if in categories 3,4,5 (low SES)

Inspection of the individual profiles indicates that overall, children with cleft palate performed within normal limits (100+/15) on the individual tests. Not all subjects performed within normal limits on each subtest. A closer look at the individual data (see table 8) indicates the following number of children who fell below a standard score of 85 on a given subtest: TOPA (S# 7, 9, 17), Rhyming Discrimination (S# 6, 9, 18), Blending Syllables (S# 7, 9, 15, 17), Blending: Phoneme (S# 7, 17), Letter Identification (S# 17).

Total raw scores for the TOLD are not reported because the individual subtests range in the number of items per subtest and the distribution of age-expectancy scores. In an effort to estimate whether a production-comprehension gap exists, raw scores were taken from two subtests (Picture Vocabulary task, considered a measure of comprehension, and Sentence Imitation task, a production measure) which were equal in the number of items (30) and had equal distributions of age-expectancy scores for ages 5 to 7. Descriptive statistics revealed that mean scores from the above mentioned subtests were as follows: Picture Vocabulary (mean = 17.20 , SD= 4.5), Sentence Imitation (mean= 18.75, SD =5.09). T-tests revealed no significant differences between scores (p=.32). This result, which indicates no difference on production and comprehension measures, is consistent with the finding using the standard scores. Raw scores for the metaphonology and phonology measures are reported in Table 11.

TABLE 11. Raw Scores for the Metaphonology and Phonology Measures

TEST	Mean	SD	Minimum	Maximum
Metaphonology	46.6 [a]	8.4	32.0	60.0
PAT-3 [b]	22.8	15.62	2.0	53.0

[a] Maximum 60

[b] raw score reflects total number of errors

A descriptive analysis of the phonological sample was performed to answer the question regarding the nature of the speech disorder within this group. Table 11 shows the prevalence of the articulation errors in the two groups of children, while Table 12 shows individual results. Speech sound productions that were deemed to be related to the child's level of phonologic development (i.e. /f/ for /θ/ and gliding of liquids) were tabulated separately from errors which were related to structural factors such as missing teeth, malocclusions, anterior fistulas, and compensatory articulations (as described by Trost, 1981). Only errors which occurred three or more times within the sample were counted.

TABLE 12. Incidence (# of children) of Articulation Errors among Cleft Types

Error Type	Cleft Type		
	CPO	CLP	Totals
Lateral distortions	1/6 (16%)	8/14 (57%)	9/20 (45%)
Dental distortions	2/6 (33%)	2/14 (14%)	4/20 (20%)
Mid palatal distortions	3/6 (50%)	10/14 (71%)	13/20 (65%)
Noncompensatory developmental errors	4/6 (67%)	12/14 (86%)	16/20 (80%)
Compensatory	0/6 (0%)	2/14 (14%)	2/20 (10%)
Audible nasal air emission	1/6 (16%)	2/14 (14%)	3/20 (15%)

The descriptive analysis revealed that overall, the majority of errors were either developmental in nature, or related to structural factors. In terms of cleft type differences, the trend indicated that the Cleft-Lip and Palate (CLP) group produced more structural related errors than their Cleft Palate only (CPO) counterparts. Of the three children with CPO who produced structurally related errors, one had undergone fistula repair in the last 9 months, while the another, who made midpalatal distortions, had an unrepaired anterior fistula. Approximately 75% (15/20) of the children had dental or occlusal problems which would have been judged to influence speech production. All of these children with occlusion problems produced at least one type of structurally-related error.

Table 13. Individual Articulation/Phonology Results

Subjects	Developmental Errors	Lateral Distortions	Midpalatal Distortions	Dental Distortions	Compensatory Articulations	Audible Nasal Air Escape	Metaphonology Difficulties[a]	Structure[b]
S1	♦							
S2	♦		♦					
S3		♦	♦					
S4								
S5	♦	♦						
S6	♦							
S7	♦	♦					♦	
S8	♦		♦			♦		♦
S9	♦		♦			♦	♦	
S10	♦		♦			♦		♦
S11	♦							
S12	♦			♦				
S13		♦	♦	♦				
S14	♦	♦	♦					♦
S15	♦	♦						♦
S16		♦	♦					
S17	♦		♦		♦		♦	
S18	♦	♦	♦					
S19	♦	♦			♦			
S20	♦	♦	♦					

[a] Results are recorded if subjects had 2 subtest scores which fell one standard deviation below the mean.
[b] Results for the structure variable are recorded if there were more than three abnormal signs on the oral mechanism exam.

Multiple Regression

Question# 3: Is there an interaction between environmental factors, IQ, structural integrity, metaphonology, phonology, and performance on language tasks?

Null Hypothesis: There will be no relationship between environmental factors, IQ, structural integrity, phonology, and language measures.

Multiple regression analysis was used to evaluate the combined effect of mother's education, IQ, structural integrity (as a measure of physical structure) and phonology (as a measure of communication experience) on their language performance. The following variables were entered into the equation, however they did not indicate predictive power: PAT-3 (p=.79), TONI (p=.76), Structure (p=.75), and Mother's education (p=.40). The remaining variables had no further predictive power.

Question#4: Is there an interaction between environmental factors, IQ, structural integrity, language, phonology, and performance on metaphonology tasks?

Null Hypothesis: There will be no relationship between environmental factors, IQ, structural integrity, language and phonology and metaphonology measures.

The relative contributions of productive and comprehensive language performance, IQ, mother's education, phonology and structural integrity in predicting metaphonological ability, as tested by a multiple regression analysis, indicated that the TONI accounted for 52 percent of the Metaphonology variance. The PAT-3 scores accounted for 15 percent of the variance in the metaphonology task, after controlling for IQ, and the production quotient accounted for a further 3 percent of the variance; however this was not significant. Table 14 shows regression coefficients and their significance levels following each step.

TABLE 14. Regression coefficient values

Step	Variables Entered	r^2	Change in r^2	Significance of change
1	TONI (1990, 1997)	0.52	0.52	p=0.0004
2	PAT-3 (1997)	0.68	0.16	p=0.01
3	Production Quotient: TOLD:P-3 (1997)	0.71	0.03	p=.24

Surgery Age

QUESTION# 5 Does early palatal repair influence speech, language and metaphonological outcomes?

Null Hypothesis: The correlation between early palatal repair and speech and language outcomes is zero.

The next analysis addressed the strength of the relationship between age of primary palatal surgery and articulation ability as measured by the PAT-3. A Spearman rank order correlational value of -.61 (p=.005) indicated a moderate and significant relationship between these variables.

Residual Structural Problems

QUESTION # 6 Do residual structural problems influence speech, language and metaphonological outcomes?

Null Hypothesis: The correlation between structural problems and speech and language outcomes is zero.

A Spearman (r) correlation coefficient was used to address the potential relationship between residual structural problems and speech and language measures. There were no significant correlations between the structure variable (a composite score which included

the results from the oral mechanism examination and perceptual measures of resonance and voice) and the TOLD: P-3 (.01), Metaphonology (.05) and PAT-3 (.23) scores. Five children required secondary management of their velopharyngeal port, an indication of residual structural problems. While it was not possible to perform a t-test between groups due to unequal group size, descriptive statistics indicated that the children who required secondary surgery had more errors on the PAT-3 (mean =37.2, SD 11.5) and subsequently had lower standard scores (mean =60.2, SD =.44) than those who did not require secondary surgery-(n=15, mean PAT-3 raw score= 16.8, SD= 15, StS mean= 86.1, SD =20.)

Cleft Type Differences

QUESTION# 7: Do children with cleft palate only perform worse on standardized measures than children with cleft lip and palate?

Null Hypothesis: There will be no difference in test results in children with cleft palate only and cleft lip and palate.

Since there has been controversy in the literature surrounding cleft type differences, the data from the Cleft Palate Only (CPO) and the Cleft Lip and Palate (CLP) group were compared using a t-test. (See table 15). The only significant difference between groups was in terms of raw phonological scores (# of errors) (t=-2.11, df=18, p=<.04). See table 12 for a detailed description of the types of errors made by each group.

TABLE 15. Means and Standard Deviations for CP0 (n=6) and CLP (n=14) Groups on Phonology, Metaphonology and Language measures.

	CPO		CLP	
	Mean	SD	Mean	SD
TOLD	109.33	9.31	112.86	11.59
METAPHONOLOGY Raw score	48.33	5.68	45.79	9.43
METAPHONOLOGY Standard Score	104.19	5.45	98.27	8.25
PAT-3 Raw	12.59	10.56	27.29	15.59
PAT-3 Standard Score	87.33	19.04	71.71	17.85

SUMMARY

Morphosyntax and Vocabulary Ability

In general, children with cleft palate scored within normal limits on all TOLD:P-3 Subtests. There was no difference between comprehension and production scores, nor was there a difference between cleft types. There was no correlation between the structural integrity of the oral mechanism or the age of primary palatal surgery and language outcomes.

Metaphonological Ability

Children with cleft palate scored within normal limits on the metaphonology tasks; however, there was individual variation noted. There was no correlation between the structural integrity of the oral mechanism or the age of primary palatal surgery and metaphonology outcomes. There was no difference between cleft-type groups. A multiple regression analysis revealed that nonverbal IQ (as measured by the TONI, 1990, 1997)

accounted for 52% of the variance while the PAT-3 (1997) accounted for 15% of the variance in the metaphonology task.

Phonological Ability

Phonological ability, as measured by the PAT-3 (1997) standard scores, indicated performance below normal limits. A descriptive analysis revealed that the majority of errors made were either developmental in nature or related to structural factors. In terms of cleft type differences, the trend indicated that children with cleft lip and palate produced more structurally related errors than their cleft palate only counterparts. A moderate correlation (-.61) was noted between age of primary palatal surgery and PAT-3 standard scores. No correlation between phonology and metaphonology (.26) scores was noted. However, in a multiple regression analysis, phonology accounted for 15% of the variance in the metaphonology task, once IQ was accounted for, the most significant variable.

CHAPTER FOUR: DISCUSSION

The purpose of this investigation was to examine the phonology, metaphonology and language skills in children with cleft lip and palate. Standardized tests were administered to twenty children with isolated cleft lip and palate and cleft palate only.

LANGUAGE ABILITY: MORPHO-SYNTAX AND VOCABULARY

The major finding was that children with cleft palate demonstrated language skills which were within normal limits when measured by the Test of Language Development-Primary (TOLD:P-3, 1997). In addition there was no difference between language production and comprehension abilities as measured by the Speaking and Listening Quotients. These finding are not consistent with past research which has documented language delay from birth to school age years (e.g. Dalston, 1990; Fox et al. 1978; Jocelyn et al., 1996; Lynch, 1986; Nation, 1970; Eliason & Richman , 1984; Smith & McWilliams, 1968). A brief re-examination of these studies might help to account for these contradictory findings considering the following issues: (1) cleft type differences, (2) subject selection (3) environmental factors

For the most part, the body of literature outlining language development in children with cleft palate fails to report cleft-type differences (Dalston, 1990; Nation, 1970; Fox et al., 1978;). Given the fact that children with cleft palate only may be more at risk than their cleft lip and palate counterparts for having an undetected syndrome associated with cognitive delay (e.g., Velo-Cardio Facial syndrome), knowing the breakdown of cleft type performance is essential to determine the nature of language delay. The findings of this study indicated no difference in performance between cleft types on the language measure. This finding was expected given that an attempt was made to exclude subjects who had syndromes associated with cognitive delay.

Given the recent advances in medical genetics, it is now possible to detect more of these syndromes than was previously possible (Shprintzen & Goldberg, 1995). Therefore this

study was motivated to improve the subject selection process and to report the language abilities of children without known syndromes. The subjects all had normal nonverbal skills as measured by the Test of Non-Verbal Intelligence (TONI, 1990, 1997). The findings indicated that when children with syndromes associated with cognitive delay are excluded, children with isolated cleft palate and cleft lip and palate can have language abilities that are within normal limits on the TOLD: P-3 (1997).

Previous studies which report language delay within this population have examined environmental factors such as SES, length of hospitalization, hearing loss, etc. (Nation, 1970; Fox et al., 1978; Jocelyn et al. 1996). Many have failed to control for factors such as mother's education and family history of speech and language problems, which may contribute to the perception of the language delay in this population. This study collected data on family history of speech and language difficulties and found that 15% of the sample (3/20) indicated positive family history. A closer inspection of the data revealed that the two children who had the lowest TOLD Quotient scores (S#17 & 19) had a positive family history of speech and language difficulties. To evaluate the effect of mother's education on the language performance measure, it was added as a variable into the multiple regression equation; however, it did not further the predictive power.

Given the cross-sectional nature of this study, another possible interpretation of the results could be that these children, at age 5 and 6, are in a developmental plateau. Scarborough and Dobrich (1990) describe this period as an "illusionary recovery" because these children appear to be developing within normal limits however they may go on to have future difficulties with oral language and reading.

Another interpretation of the results could be attributed to the fact that parents of children with cleft palate have an increased awareness of speech and language and hearing development. Parents are given this information by the speech pathologist on the cleft palate team and are immediately referred to the local speech language pathologist for monitoring. This

is close to optimum in terms of the management of both hearing and speech-language development.

LANGUAGE ABILITY: METAPHONOLOGY

This study adds to the literature because it documents the metaphonological abilities of children with cleft palate. The results indicated that children with cleft palate can score within normal limits on tests of phonological awareness. In addition, results from the digit span task suggest that children with cleft palate can have adequate verbal working memory to perform metaphonological tasks. These findings rule out the possibility that this group of subjects had underlying speech processing difficulties which could contribute to later reading difficulties. Given their performance on the auditory word discrimination subtest on the TOLD: P-3 (1997), it appears that children with cleft palate can establish secure and stable phonological representations despite structural deficiencies.

The findings of the multiple regression analysis indicated that the TONI (1990, 1997) scores accounted for 52% of the variance on the metaphonology measure, while the PAT-3 (1997) scores accounted for 15% of the variance. An examination of the nature of the tasks involved in the TONI might explain its significant contribution to the variance in the metaphonology measure. The TONI consisted of tasks which involve abstract reasoning and problem solving. One might argue that phonological awareness tasks involve similar sets of skills. The contributions of the PAT-3 scores will be discussed in the following section.

LANGUAGE ABILITY: PHONOLOGY

Given the claims that phonological impairment may hinder the establishment of secure and stable phonological representations (Webster et al.,1997) and the reports of phonological disorders within the cleft palate population (Chapman, 1993), there was a need to document the nature of the speech disorder. To determine the nature of the speech disorder this study examined the types of errors produced. The results indicated that for 90% of the children, the errors reflected deficits in the structural integrity of the oral mechanism, not phonological

representation problems. These results appear to be consistent with the findings of Chapman (1993) which indicated that despite phonological delay at age 3, by age 5 children with cleft palate were performing similar to their non-cleft counterparts. A closer inspection of the profiles of the two children who continued to use compensatory articulation (S#17 & 19) indicated that they had TOLD Quotient and Metaphonology scores at the lower range of the group (although still within normal limits). Both of these children had average TONI and Digit Span scores. In addition one of these children had a positive family history of speech and language difficulties.

Dental and occlusal problems appeared to be the primary causes of articulatory errors in this subject group. Midpalatal and lateral distortions were the most common errors. It is difficult to determine whether dental or occlusal problems alone caused these errors or whether it was a combination of both problems. Missing teeth and malocclusions can both contribute to distortions with sibilants and affricates because they interfere with normal tongue positioning. Missing teeth distort sounds because the tongue moves forward to fill the space of the missing teeth while malocclusions of the maxillary and mandibulary arches prevent the tongue from acquiring a normal resting position, thus changing the place of articulation and interfering with the grooving aspect of the tongue. Unfortunately data on mouth breathing patterns of these children was not collected; therefore one cannot rule out the possibility that mouth breathing inhibited maxillary development and contributed to subsequent articulatory errors.

RELATIONSHIPS BETWEEN PRIMARY REPAIR, PHONOLOGY AND METAPHONOLOGY

The finding of a moderate correlation between age of primary palatal repair and PAT-3 (1997) scores raises several issues regarding the nature of early phonological acquisition in this population: To what extent does an inadequate oral mechanism interfere with babbling and the subsequent lexical acquisition, and what role does early conductive hearing loss play in development? Prior to primary palatal repair these children have inadequate velopharyngeal

mechanisms but unlike children with tracheostomies (Oller, Levine, Cobo-Lewis, Eilers & Pearson, 1998), children with cleft palate are not inhibited from vocalizing nor are they prevented from exercising the vocal tract (moving the tongue, opening and closing the mouth). O'Gara and Logemann (1990) investigated the development of early phonetic inventories of children with cleft palate and found that the "structural constraint of an unrepaired cleft palate during the prelinguisitc period could delay the early use of the high intraoral pressure target, particularly /p/, /b/, /t/, /k/ and /g/" (pg.718). The subjects in the O'Gara and Logemann (1990) study included 23 infants who were followed from 3 to 36 months of age, and whose mean age of primary palatal surgery was 9.3 months. The authors suggested that compensatory articulations such as glottal stops and pharyngeal fricatives may have their origins in babbling. In addition they concluded that children with cleft palate would benefit from early speech and language intervention prior to primary palatal repair. The authors claimed that "enrichment of the phonetic features of variegated babbling prior to primary palate repair and shaping of acceptable orally targeted consonants during the early months after palatoplasty may be important to later, more normal, phonemic development of oral articulations" (pg. 720). Estrem and Broen (1989) investigated the early speech production of five children with cleft palate and found that their lexicons contained fewer words beginning with [-sonorant] phonemes and more words beginning with [+sonorant] phonemes. In addition their lexicons contained more labial and glottal sounds than did the control group of non-cleft children.

The findings of the current study seem to suggest that early palatal repair is related to better speech outcomes at 5 to 6 years of age. The mean age of primary palatal surgery in this group was 10.10 with a range from 7 to 12 months; thus it appears that age of surgery plays a role despite the relatively small age range. This finding might suggest that children who have surgery prior to the onset of their first word and at an earlier stage of babbling have better outcomes. Perhaps this is because earlier surgery allows the child to practice articulating sequences of sounds that resemble the targets with a functional mechanism. One must interpret

these results with caution because this was a retrospective study and no information was provided regarding the early productions of these children. It is not furthermore possible to determine the role that aggressive hearing management played in the development of the early phonetic repertoire. In light of O'Gara and Logemann's work (1990), all parents were advised about the importance identifying compensatory articulations in the infant's early phonetic repertorire, and intervention was initiated in an attempt to prevent the firm establishment of these errors in the phonological system. Given these factors it is not possible to attribute the better speech outcomes to early surgery alone.

While there appears to be a link between early palatal surgery and performance on the PAT-3, there is not a direct link between age of surgery and performance on metaphonology tasks. However, the multiple regression analysis indicated that the PAT-3 scores accounted for 15% of the variance on the metaphonology measure. This seems to suggest that there may be an indirect link between earlier palatal surgery and later metaphonology difficulties, which is mediated by phonological skills. But questions remain regarding the nature of the phonology scores themselves. Perhaps the phonology scores reflect two sorts of errors: phonological errors which are related to metaphonology and articulation errors which are related to the age of surgery. In an attempt to answer this question the PAT-3 (1997) was rescored. All distortions were counted as correct if there was a different distortion for each phonemic target. The revised scores may better reflect the phonological abilities, while the initial scores may reflect the combination of phonological and articulation abilities. Table 16 notes the revised scores overall and by cleft type.

Table 16. Revised Phonology Scores by Cleft Type

	Mean	SD	Minimum	Maximum
Overall	88.1	15.82	60.00	114.00
Cleft Lip and Palate	85.00	16.2	60.00	114.00
Cleft Palate Only	95.33	13.62	68.00	105.00

Correlations between the age of surgery and the revised PAT were calculated; however, no significant correlation was found (-.36, p=.39). This result confirms the hypothesis that age of surgery is related to articulation outcomes rather than phonological outcomes. When the revised PAT-3 (1997) score was entered into the multiple regression analysis it did not account for a significant amount of the variance in the metaphonology measure. The fact that the articulation and phonology measure accounted for 15% of the variance, while the phonology measure did not provide any predictive power invites a theoretical discussion. If one takes an interactive activation approach to language acquisition, which posits an interaction among language units, then it is possible to suggest that a combination of articulatory and phonological problems may lead to inappropriate activation of phonological units within the network. This model would also predict that if children with cleft palate have phonological deficits they may also be at risk for morphosyntactic deficits. While the performance within normal limits on morpho-syntax and vocabulary measures appears to be in conflict with the interactive activation model, one might argue that severity of the disorder plays a role. Larrivee and Catts (1999) suggested that children who had more severe phonological disorders performed significantly worse on phonological awareness and language measures. In this case, the majority of the subjects were not considered to have severe phonological problems which interfered with intelligibility. Another possible explanation for the fact that the revised phonology measure did not account for variance in the metaphonology measure, could be

statistically based. Perhaps there were too many children near the ceiling, and an increase of subjects would create more variance.

Limitations

This study used several standardized measures to assess language ability which were administered in a standardized fashion according to the procedures found in the manual. Unfortunately due to time and budget constraints it was not possible to compare results of the cleft palate group to a control group of local peers matched for chronological age. Instead, indirect comparisons were made to a remote normative group of "peers." Another problem associated with the use of standardized tests is that they are a form of static assessment. Many supporters of dynamic assessment have argued that static assessments often fail to capture they child's ability to change or benefit from instruction.

In addition to the problems associated with using standardized tests and not having a control group, interpretation of the results are limited due to the small sample size. The sample size was limited by the following factors: exclusionary criteria, distance from testing center and the limited age range. To increase sample size in the future, studies could combine samples from different centers (although this suggestion might add additional variance as centers may differ regarding age of surgery and otitis media management).

It was not possible to collect prospective hearing data due to budget and time constraints. These data would have been beneficial because it would help to determine whether hearing loss associated with otitis media influences the language development of children with cleft palate. In addition it would allow us to make outcome statements regarding the effectiveness of otitis media management with this population.

Implications for Management/Instruction

Previous studies which report language delay in this population have failed to exclude subjects with syndromes associated with cognitive delay. The fact that no delay was found in this current investigation could be attributed to the fact that a strict subject selection process was employed. The difference in findings highlights the importance of identifying syndromes through genetic counselling. A correct diagnosis can lead to better patient management and care.

Continued orthodontic treatment is recommended for this population. It is not clear whether orthodontic treatments alone will reduce articulatory distortions because some children may have difficulties adjusting to their new occlusal closure and may not be able to change habituated patterns of articulation. Continued monitoring by the speech pathologist and orthodontist is recommended. Some children who have difficulties adjusting to new patterns of closure might benefit from visual feedback such as electropalatography.

The finding of normal language development within this population does not negate the importance of early speech and language intervention/monitoring. In fact, it may support the need for early intervention because 90% of these children had speech and language services.

Future Research

The results of this study have provided some insights into the development of language, metaphonology and phonology skills of children with isolated cleft lip and palate.

There is a need for longitudinal studies with larger sample sizes and control groups which follow children from birth to late primary. Gathering prospective hearing data and data regarding the management of otitis media would help to understand the relationship between hearing status and language development in this population.

Summary and Conclusion

The results of this study indicate that children with isolated cleft lip and palate and cleft palate only have normal morphosyntactic, vocabulary and metaphonology skills when measured with standardized language tests. Residual speech production difficulties appear to be related to dental issues such as malocclusions and missing teeth, for most children.

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APPENDIX A

TEST DESCRIPTION

LANGUAGE MEASURE

TEST OF LANGUAGE DEVELOPMENT- Primary (TOLD-P:3):

Picture Vocabulary: Children were asked to point to one of four pictures which best depicted an orally presented word (i.e., show me "dog").

Relational Vocabulary: Children were asked to orally express the relationship between two words (i.e., How are a pen and a pencil alike?) .

Oral Vocabulary: Children were asked to give oral definitions of common English words (i.e., What's an apple?).

Grammatic Understanding: Children were asked to choose one of the three pictures that best depicted an orally presented sentence. This test evaluates the ability to understand different grammatical constructions, such as negation (e.g., She can't find her ball) and relative clause (e.g., the picture that was drawn by the artist is finished).

Sentence Imitation: Children were required to repeat a sentence. Sentences increased in length and complexity.

Grammatic Completion: Children were required to complete a sentence by providing the last word. This test evaluates the ability to produce grammatical forms such as regular and irregular plurals (e.g., dresses and mice), verb endings etc.

Word Discrimination: Children were asked to say whether two orally presented words were the same or different (e.g., witch-watch).

NONVERBAL MEASURE

TEST OF NON-VERBAL INTELLIGENCE- TONI-2, TONI-3

The test consists of a series of incomplete patterns, each with six single designs of which one will correctly complete the pattern. Children were required to choose one of the six pictures which completed the pattern. Pictures in a set shared some similar properties such as size, shape and number.

SOUND AWARENESS MEASURES

THE TEST OF PHONOLOGICAL AWARENESS (TOPA)

Kindergarten Version:

Initial Sound- Same: Children were asked to look at the first picture and then chose one of three pictures which begins with the same sound as the initial picture. Children were given the

instructions orally (i.e., "The first picture is leg. The other pictures are lamp, hand and fish. Choose the one that begins with the same sound as leg.")

Initial Sound- Different: Children were asked to look at four pictures and to choose which picture has a different first sound than the other three. The instructions were orally presented (i.e., "Look at the pictures fork, fan, foot, shirt. Choose the one that has a different first sound than the other three.").

Early Elementary Version:

Ending Sound-Same: Children were asked to look at the first picture and then chose one of three pictures which ends with the same sound as the initial picture. Children were given the instructions orally (i.e., "The first picture is ball. The other pictures are smile, horn and duck. Choose the one that ends with the same sound as ball.")

Ending Sound- Different: Children were asked to look at four pictures and to choose which picture has a different first ending than the other three. The instructions were orally presented (i.e., "Look at the pictures: doll, rain, nail, smile. Choose the one that has a different last sound than the other three.").

THE PHONOLOGICAL AWARENESS TEST

Rhyming-Discrimination: Children were required to say whether two orally presented words rhymed (i.e., "Do these words rhyme? book-look).

Rhyming-Production: Children were required to produce rhymes (i.e., "Tell me a word that rhymes with can"). Nonsense rhyming words were acceptable.

Blending- Syllables: Children were orally presented the parts of a word and were required to guess the target word (i.e., "ta-ble, What word is this?").

Blending-Phonemes: Children were orally presented the sounds of a word and were required to guess the target word (i.e., "p-o-p, what word is this?").

Letter Identification- Consonants: Children were shown a series of letters and they were asked to provide the name of the letter and it's corresponding sound (i.e., "This is the letter t and it makes the 'tuh' sound").

MEMORY MEASURES

WECHSLER INTELLIGENCE SCALE FOR CHILDREN-III (WISC-III):

Digit Span: Children were asked to repeat sets of numbers, as presented, or in reverse order.

TEST OF LANGUAGE DEVELOPMENT- Primary (TOLD-P:3):

Sentence Imitation: Children were required to repeat a sentence. Sentences increased in length and complexity.

SPEECH MEASURE

PHOTO ARTICULATION TEST-3(PAT-3):

Children were asked to look at the pictures and provide the appropriate labels (i.e., "I'm going to show you some pictures and I want you to tell me what they are"). Their utterances were recorded on an audiotape and an analysis or their phonology was performed at a later time. Errors were classified as misarticulations ("f" for "th" as is "fumb" for "thumb") or distortions ("nailsth")

APPENDIX B

Richmond Health Department: Screening and Referral Procedure

1. The child, tester and equipment should be arranged in such a way that the tester can observe the child's non-verbal response to sound but the child cannot see the tester's hand on the interrupter switch.

2. Before putting the earphones on the child's ear, explain and demonstrate the procedure.

3. Procedure #1 (Normal Procedure)

RIGHT EAR	Phase A	1000 Hz 2 second sound at 40 dB (Orientation)
		1000 Hz 2 second sound at 40 dB (Orientation)
	Phase B	1000 Hz 2 second sound at 20 dB (Do not test below)
		2000 Hz 2 second sound at 30 dB
		2000 Hz 2 second sound at 20 dB (Do not test below)
		4000 Hz 2 second sound at 35 dB
		4000 Hz 2 second sound at 25 dB (Do not test below)
		500 Hz 2 second sound at 35 dB
		500 Hz 2 second sound at 25 dB (Do not test below)
LEFT EAR		1000 Hz 2 second sound at 30 dB
		1000 Hz 2 second sound at 20 dB (Do not test below)
		2000 Hz 2 second sound at 30 dB
		2000 Hz 2 second sound at 20 dB (Do not test below)
		4000 Hz 2 second sound at 35 dB
		4000 Hz 2 second sound at 25 dB (Do not test below)
		500 Hz 2 second sound at 35 dB
		500 Hz 2 second sound at 25 dB (Do not test below)

If child does not respond at any point during the test (after orientation) stop the test. Child failed. Retest same day. Record results. Refer to audiologist if failed twice.

APPENDIX C

TEST ORDER

TEST ORDER ONE	TEST ORDER TWO	TEST ORDER THREE
TONI- (1990,1997)	PAT-3	Hearing Screening
TOLD (1997)	WISC	TOLD
TOPA (1994)	Phono Awareness Test	Phono Awareness Test
PAT-3 (1997)	Hearing Screening	O.M.E.
WISC (1991)	O.M.E.	TONI
Phonological Awareness Test (1995)	TOLD	PAT-3
Hearing Screening	TONI	TOPA
O.M.E.	TOPA	WISC