PHONOLOGICAL AWARENESS, SPEECH, AND LANGUAGE SKILLS IN CHILDREN WITH CLEFTS

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ABSTRACT

The purpose of this study was to examine the speech, language, and phonological awareness skills of children with cleft palate ranging in age from 4 to 6 years old. Twenty-three children with clefts were compared to a sample of 23 matched peers without clefts on standardized measures of phonological awareness, speech production, and language skill development.

Paired samples t-tests did not yield statistically significant between group differences on measures of phonological awareness. Similarly, comparison of children with clefts to normative data available with phonological awareness testing measures also did not yield statistically significant differences.

Further examination of the phonological awareness skills of children with clefts was conducted with regard to speech production, language skills, and hearing status. Correlational analyses revealed a positive relationship between speech production and alliteration awareness. Two measures of receptive language were found to be positively correlated to alliteration awareness and rhyme awareness. Finally, there was a positive relationship between alliteration awareness and the number of recent ear infections a child had experienced.

The results of this study suggest children with clefts demonstrate phonological awareness skills that are similar to their peers. Given the relationship identified between phonological awareness skills and receptive language skills, practitioners working with

children with clefts should not only continue to monitor the speech production skills of children with clefts, but also assess language development and phonological awareness in order to ensure these children have the skills to become strong readers.

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CHAPTER 1

Introduction

The aim of this investigation was to explore the speech, language and phonological awareness skills of young children with clefts ages 4 to 6 years old. Despite the fact that children with clefts are often characterized by severely impaired speech production through the preschool years and have been shown to be at risk for speech, language, and reading difficulties into elementary school, the literature has not examined the role of phonological awareness in this population. Instead, research relating to children with clefts typically focuses on specific aspects of speech production, and only recently, has it begun to focus increasingly on language and reading skills. This investigation applies what is known about the contributions of phonological awareness, speech production and language skills to reading development and examines these skills in children with clefts.

Phonological Awareness and Literacy Development

Phonological awareness is “the awareness of sound structure of spoken words” (Gillon, 2004, p. 2). As of late, phonological awareness has taken center stage in both the reading and speech and language literature, perhaps due to the fact that it provides a link between oral and written language. The reading and written language literature are replete with research regarding the role of phonological awareness in reading development in the general population (see e.g., Bishop, 2003; Hecht, Burgess, Torgesen, Wagner, & Rashotte, 2000; Kirby, Rauno, & Pfeiffer, 2003). The first reports of the causal role phonological awareness plays in reading development appeared in the late 1980’s (Wagner & Torgesen, 1987), and such evidence continues to be reported today (Anthony & Lonigan, 2004; Engen & Hoien, 2002; Kamii & Manning, 2002; Torgesen, Wagner & Rashotte, 1994). At the same time, the last decade has given rise to a dramatic increase in research examining the efficacy of interventions that focus on phonological awareness (Byrne, Fielding-Barnsley, & Ashley, 2000; Ehri et al., 2001; Hurford et al., 1994), and classroom strategies that facilitate the development of phonological awareness skills (Wasik, 2001; Yopp & Yopp, 2000). Given this emphasis on the importance of phonological awareness skills, it is critical to explore the relationships between phonological awareness and aspects of oral communication, such as speech production and language skill development, as they relate to literacy.

Phonological Awareness and Speech Disorders

Several studies have looked at the phonological skills of children with speech impairments. Children with expressive phonological impairments have poorer phonological awareness skills than their typically developing peers (Bird, Bishop, and Freeman, 1995; Raitano, Pennington, Tunick, Boada, & Shriberg, 2004). Additionally, in cases where the speech production issues have resolved, deficits in reading and spelling might remain (Bird et al.; Raitano et. al). Severity of speech and language deficits is also a critical factor to consider, as children at greater risk for reading impairment are known to have the most significant deficits in terms of phonological awareness, language skills, and speech productions (Larrivee & Catts, 1999). For these reasons, children who begin elementary school with impaired speech productions and even those who had speech delays that have resolved by the time they begin to read are at risk for reading difficulties.

Phonological Awareness and Language Disorders

Oral language skills are often related to deficits in reading skill development (Catts, 1993, 1996; Catts & Hogan, 2003; Snyder & Downey, 1997). Deficits in the understanding and use of morphological markers, syntactic rules, and vocabulary lead to significant deficits in terms of written language comprehension. In addition, phonological awareness skills have been shown to be related to language skill development at different stages of reading development (Cooper, Roth, Speece, & Schatschneider, 2002; Rvachew, 2006; Stothard, Snowling, Bishop, Chipchase & Kaplan, 1998). Deficits in phonological awareness impact reading and spelling skills, as the child may be unable to understand how sounds in spoken words and letters in written words come together. Language skills, including phonological awareness skills, are therefore critical to reading comprehension.

Speech and Language Skills of Children with Clefts

Researchers have documented differences in early speech development between children with clefts and typically developing children. Differences in sound productions have been noted in children with clefts from infancy (Chapman, 1991; O'Gara & Logemann, 1988) through the school-age years (Lowe & Scherer, 2002). These errors may be due to structural limitations, learned behaviors that compensate for existing or repaired structural limitations, and even developmental errors. While the goal of practitioners working with children with clefts is to achieve speech production skills that are within age expectations by the time a child enters kindergarten, many children with clefts continue to demonstrate speech errors well into their school age years.

Although most research investigating communication skills in children with clefts focuses on speech production, a limited number of investigations have explored language development in this population. Infants and toddlers with clefts are at risk for decreased language skills when compared to their peers (Broen, Devers, Doyle, Prouty, & Moller, 1998; Jocelyn, Penko, Rode, 1996; Neiman & Savage, 1997; Speltz et al., 2000). Studies examining language skills in the preschool years continue to show differences between children with clefts and their typically developing peers. Differences have been noted in vocabulary (Brennan & Cullinan, 1974; Nation, 1970), expressive language skills only (Spriestersbach, Darley, & Morris, 1958), and overall receptive and expressive language skills (Philips & Harrison, 1969; Smith & McWilliams, 1968). However, most studies indicate that children with clefts have language skills that are within normal limits; this finding often leads practitioners to focus only on speech production skills when addressing the communication needs of children with clefts. As demonstrated by these studies, children with clefts may exhibit delays or differences in language development that emerge in the infant and toddler period and remain well into the preschool and school age years.

An important factor that is often associated with delayed communication skills in children with clefts is their struggle with middle ear dysfunction. Individuals with cleft palate have abnormal functioning of the Eustachian tube, causing fluid build up in the middle ear. This is associated with mild conductive hearing loss, which could lead to permanent hearing loss (Kummer, 2001). Despite preventative measures such as insertion of pressure equalization tubes, individuals with clefts often have ongoing difficulty with middle ear infections into their school age years, placing them at risk for decreased language skills and speech delays, which can in turn impact a child's early reading abilities.

Literacy Development of Children with Clefts

Children with clefts are at risk for developing reading difficulties beyond what is expected in the general population (Richman, Eliason, & Lindgren, 1988). Despite this, only one study has attempted to describe the type of reading disability seen in children with clefts. Although the evidence presented herein highlights the interactions between speech, language, and phonological awareness skills in the general population, there has yet to be a study that has examined early phonological awareness development in children with clefts or areas of reading beyond the phoneme and individual word level. Given the association between speech impairment and phonological awareness, it is important to explore the potential relationship between speech impairments and phonological awareness skills in children with clefts. Additionally, weaknesses in language development may inhibit a child's ability to become a skilled reader. These issues have not been fully explored as they relate to children with clefts.

Summary

In conclusion, strong phonological skills have been linked to positive reading outcomes. Speech production and language skill development have been shown to be related to phonological awareness skills such that deficits in either of these areas have been associated with decreased phonological awareness skills. Children with clefts are often characterized by severely impaired speech production through the preschool years and have been shown to be at risk for oral language and reading difficulties into elementary school.

This investigation examines the role of phonological awareness in the continued study of oral and written communication skills of individuals with clefts. The current investigation not only contributes to the discussion of speech production and oral language skills in preschool and early school-age children with clefts, but also examines the relationship between these skills and phonological awareness in children with clefts. Additionally, this investigation endeavors to explore the relationship between hearing status and measures of phonological awareness in children with clefts. The information gained from this study will enable speech-language pathologists and other members of the Cleft Palate Team to target assessments and interventions in a manner that supports strong oral and written communication skills of children with clefts.

CHAPTER 2

Review of the Literature

Because very little is known about the phonological awareness skills of children with clefts, this review will describe the relationships between speech, language, and phonological awareness skills in children in the general population, while detailing what is known about each of these skill sets in children with clefts. While there is no published research specifically addressing phonological awareness skills of young children with clefts, there are a number of lines of research that converge to suggest that a more complete understanding of the phonological awareness skills of children with clefts is needed. For example, there is both theoretical and empirical evidence of a strong relationship between spoken and written language, spoken language and phonological awareness, speech production and phonological awareness, as well as written language and phonological awareness within the general population. There is also evidence that shows children with clefts are at risk for speech disorders, language delay, middle ear dysfunction, and deficits in reading skills. Each of these areas of converging evidence will be reviewed in order to demonstrate the need to investigate phonological awareness, speech, and language skills as they relate to literacy development in children with clefts.

Phonological Awareness and Literacy Development

Phonological awareness is a relatively new area of investigation, but its importance to literacy achievement is well documented in the literature. Literacy learning success is influenced by an interaction of factors that include oral language skills and phonological and phonemic awareness skills. Phonological and phonemic awareness (PPA) have received a great deal of attention in recent years because of our increasing understanding of their contribution to literacy achievement. Defined as "one's awareness of and access to the phonology of one's language" (Wagner & Torgesen, 1987, p. 192), PPA includes such skills as the ability to manipulate phonemes, syllables, and rhyming words (Ehri et al., 2001).

Phonological and phonemic awareness skills develop in progression at approximately the same time children are learning the earliest skills related to reading (Wagner & Torgesen, 1987). The category of skills defined as PPA skills vary greatly, but all are related to an individual's ability to hear and manipulate sounds in spoken language. Certain PPA skills are easier for young children than others, as it typically is easier for children to engage in tasks at the syllable level than at the phoneme level (Wagner & Torgesen, 1987). Syllable level PPA skills such as rhyming and syllable segmentation tasks are therefore early developing skills. Gillon (2004) provides a comprehensive list and description of PPA tasks and their typical order of development. (See Appendix A for the list provided by Gillon, 2004).

Generally accepted as the earliest developing PPA skill, syllable level tasks require the individual to manipulate syllables within words. Gillon (2004) identified four syllable level PPA tasks including syllable segmentation, syllable completion, syllable identity, and syllable deletion. Syllable segmentation tasks require the individual to separate words into syllables, often by counting or tapping out syllables as the word is spoken. Syllable completion tasks require an individual to complete a target word when only a portion of the word is presented auditorily in the context of a verbal cue or picture. Syllable identity tasks are those that present several words and ask the individual what parts of the words share the same sounds. Syllable deletion tasks require an individual to modify a word by deleting either the initial syllable or final syllable of the word (Gillon, 2004). Syllable level PPA skills eventually support individuals as they segment words for reading and spelling.

Rhyme awareness, a PPA skill typically developing later than syllable level skills, is the ability to isolate words at the intra-syllabic level (Gillon, 2004). Types of rhyme awareness tasks described by Gillon (2004) include spoken rhyme recognition, spoken rhyme detection, spoken rhyme generation, and onset-rime blending. Spoken rhyme recognition includes the ability to compare two words and determine whether or not they rhyme. Spoken rhyme detection tasks require the individual to determine which word in a group of several words does not rhyme with the others. Spoken rhyme generation involves the ability to come up with a word that rhymes with the others. Rhyme awareness can be assessed through these as well as other tasks that require manipulation of words at the intra-syllable level. While the order of acquisition between specific rhyme and syllable level PPA skills can be inconsistent, these skills emerge before phoneme level skills (Carroll et al., 2003), and typically develop as precursors to decoding and spelling abilities.

Phonemic awareness skills, a subset of PPA tasks, reflect an individual's ability to manipulate words at the phoneme level. Phoneme-level skills described by Gillon (2004) include the following: (a) alliteration awareness, (b) phoneme matching, (c) phoneme isolation, (d) phoneme completion, (e) phoneme blending, (f) phoneme deletion, (g) phoneme segmentation, (h) phoneme reversals, (i) phoneme manipulation, and (j) spoonerisms. Alliteration awareness tasks present several words and ask the individual to identify the word that begins with a different sound. Phoneme matching tasks provide a target word and ask the individual to identify from two or three comparison words the word that begins with the same sound as the target word. Phoneme completion tasks require an individual to complete a word when only a portion of a word is presented auditorily in the context of a verbal cue or picture. Phoneme blending tasks present individual phonemes auditorily and require the listener to put the sounds together to make a word. Phoneme deletion, or elision, requires the individual to modify a word by deleting a phoneme from the word and create a new word. Phoneme segmentation tasks require the individual to separate words into individual phonemes. Phoneme reversals require the individual to say the word backwards. Phoneme manipulation tasks present a word and then require the individual to substitute a phoneme in the first word with a separate target phoneme. Finally, spoonerisms require phoneme manipulation between words whereby a phoneme is stripped from one word and replaces a phoneme in another word. Each of these tasks reflects increasing sophistication in an individual's awareness of and ability to manipulate phonemes in spoken words. While it is unclear that the ability to demonstrate each of these skills is required for all individuals who achieve success in early reading, there is clearly a relationship between the ability to demonstrate these skills and early literacy achievement for most children (Mann, 1993; Swank & Catts, 1994).

The impact of these various PPA skills has been studied at length. Wood and Terrell (1998) investigated phonological awareness skills in 30 preschool children and followed their progress in literacy development over a two-year period. The extensive phonological awareness battery used in this study consisted of a variety of syllable level, rhyme awareness, and phoneme level tasks. Initially, the preschool children performed as would be expected, in that they performed best on syllable level tasks, then onset-rime tasks, and then phoneme level tasks. Over the two-year longitudinal portion of the study, the authors found that rhyme awareness, measured in terms of segmentation and rhyme detection tasks was a strong predictor of reading and spelling ability (Wood & Terrell, 1998). Additional investigations have focused on phoneme awareness tasks as predictors of reading ability in later grades. Mann (1993) reported that alliteration awareness skills predict 30-40% of the variance in reading abilities in first grade.

While syllable and onset-rime tasks are generally accepted as early developing skills and phoneme level tasks as later skills, it is important to remember that not all children develop these skills in the same order. One study investigated the developmental paths children take in their acquisition of both phonological awareness skills and reading skills (Christensen, 1997). Six hundred thirty children were tested on measures of phonological awareness before entering formal schooling and then at mid-year and the end of the first school year. Results suggest that while many children follow the expected developmental path, a “substantial number of children show divergence from this sequence” (Christensen, 1997, p. 352).

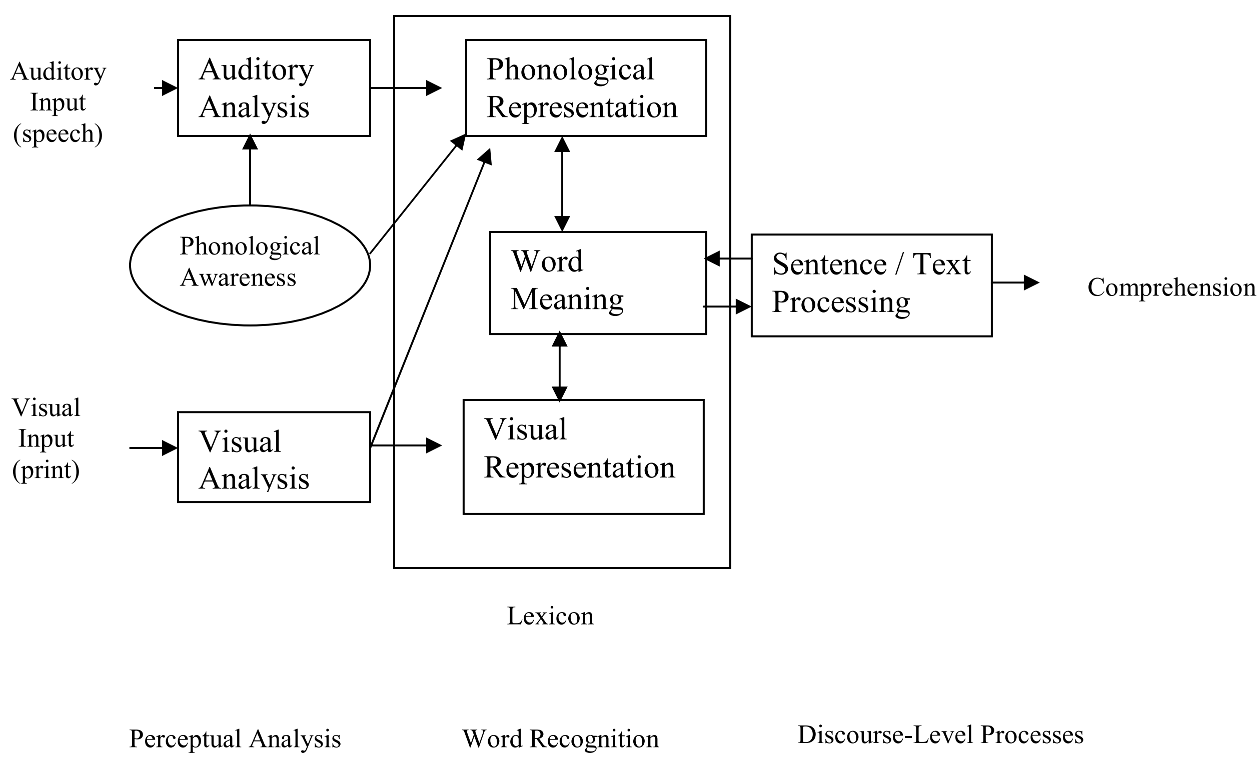
The development of phonemic awareness contributes to a child’s understanding of how sounds in spoken words and letters in written words come together, and, subsequently to the development of reading and spelling skills (Bryant, MacLean, Bradley, & Crossland, 1990; Gillon, 2004; Wagner & Torgesen, 1987; Wasik, 2001). For example, phoneme identity helps children attend to different sounds in words. The listening task of identifying “which word starts with a different sound?...cat—cup—dog?” allows the child to realize that, in addition to the meaning behind words, words are made up of sounds as well. A more sophisticated phonemic awareness skill such as phoneme segmentation helps the child attend to all of the sounds in a word; this task helps to improve spelling skills (Ehri et al., 2001).

While phonological awareness skills have been shown to help children learn to spell and decode written text, not all children who achieve high levels of phonemic awareness become good readers (Byrne, Fielding-Barnsley, & Ashley, 2000). This is important to keep in mind, as phonological awareness training does not address all of the other skills (for example, language skills such as semantics or syntax) that must come together in order to read successfully. In fact, research has demonstrated that in early elementary school, when successful reading depends heavily on successful word reading skills, decoding is more predictive of reading ability than language skill development; however, in later elementary school when successful reading requires high levels of comprehension, language skills such as semantic and syntactic knowledge are more predictive of reading skill than decoding (Swank & Catts, 1994). As we move toward a more comprehensive understanding of the relationship among phonological awareness, written language, and broader areas of language comprehension a strong theoretical framework will help demonstrate the contribution one makes to another.

A Theoretical Model of Spoken and Written Language

Various models of written language and the processes that are related and contribute to it exist (see e.g., Cunningham, 1993; Hoover & Gough, 1990). The model serving as the theoretical framework for the current investigation is that described by Kamhi and Catts (1998). It was selected because it emphasizes the relationship between spoken and written language comprehension (Kamhi & Catts, 1998). Given the hypothesized relationship between the speech impairments that are pervasive in the population of young children with clefts, a model that emphasizes the relationship between spoken and written language, rather than one that merely describes the cognitive constructs underlying written language comprehension, was critical. As illustrated in Figure 1, Kamhi and Catts describe the interactive relationship among spoken and written language comprehension that includes three major phases leading to comprehension: (a) perceptual analysis, (b) word recognition, and (c) discourse-level processes. The term *interactive* is appropriately used to explain this model since it is not unidirectional; rather, both top-down and bottom-up methods interact to lead to comprehension at various levels. In an attempt to explain the model clearly, the bottom-up path will be explained first, but it must be stressed that this model is transactional in nature with bottom-up and top-down processes developing simultaneously.

Figure 1. Adapted from Kamhi and Catts' Model of Spoken and Written Language Comprehension (1998)



Beginning at the bottom of Kamhi and Catts' model of spoken and written language, the first step of language comprehension involves perceptual analysis. With regards to oral language comprehension, perceptual analysis requires that a sound signal first be recognized as a discrete unit of speech that is a component of the sound system of the listener's language. This input requires knowledge of the phonology of language and draws upon phonological awareness skills. Perceptual analysis in written language requires that input be recognized as print comprised of letters that correspond to sounds in the reader's language. Once the input has been recognized in either oral or written language, the second phase, word recognition, begins.

There are three major components to the word recognition phase, all of which involve the lexicon, or vocabulary of the language system. These three components include phonological representation, visual representation, and word meaning. In oral language, the word recognition phase begins as the speech perceived in stage one is parsed into phonological representations of words that are accessed from the lexicon. In adults, this is accomplished at an unconscious level by analyzing the word based on its own unique pattern of phonemes, the smallest discrete sounds in a language. This process is slightly different in very young children, however, as they are not yet able to break words into individual phonemes. Instead, they "chunk" words or phrases together. For example, the common phrase *give me* as used by young children, is often represented in their lexicon as the single unit *gimme*. As children mature, and their phonological awareness develops, they develop the skills to break the speech they perceive into individual words, then syllables and phonemes. Regardless of the route through which the phonological representation is identified as a word or phrase within an individual's lexicon, the phonological representation is required before the word meaning is activated and the important syntactic and semantic rules about the word housed in the lexicon can be accessed. As described here, the path for spoken words is straightforward; written words, however, may take a slightly more complex path before the word meaning and related syntactic and semantic rules are accessed.

When a written word has been perceived as a word that is potentially within one's language, it may be recognized immediately. In this case, the visual representation of a word has been stored in the lexicon as a unit, and the meaning is accessed based on the visual representation alone. This process is often referred to as the direct or visual approach (Kamhi & Catts, 1998). However, there are instances when a reader encounters an unfamiliar word that is not immediately recognized. When this occurs, the individual can attempt to identify the word using the phonological representation of the word. This requires an individual to "sound out" the word using knowledge of phoneme-letter or syllable-sound correspondence to map individual or groups of phonemes and blend them together until they resemble a symbol that is housed in the lexicon. This, according to Kamhi and Catts, is the phonetic approach to reading. This process of accessing a phonological representation of a word that was originally presented visually requires strong PPA skills.

The discourse-level processing that occurs in stage 3 begins after the word meaning has been accessed from the lexicon. Discourse processing involves understanding sentences, conversations, and narratives beyond the single word level. It is at the level of discourse processes where oral language skills become critical. Kamhi and Catts (1998) identify three different areas of oral language that are used to support comprehension through discourse level processes. First, readers use structural knowledge of syntax and morphology to gain information about unfamiliar words. This involves not only understanding which words serve various functions (i.e. a noun versus a verb), but also requires the ability to organize words in a manner that is meaningful. Second, readers use propositional knowledge to remember the main ideas presented in text, rather than the exact words and word order that was used. With propositional knowledge, readers organize and remember concepts based on their predicates and their related arguments. Finally, world knowledge, which comes from knowledge of facts as well as interpersonal relationships, allows readers to determine if what they hear or read makes sense. These three levels of language processing interact to form meaning based on what we hear and read.

The transactional nature of this model makes it possible to explain how a word that is not recognized can be inferred from context. For example, the word *togs* is not a meaningful word to most Americans, and therefore does not trigger the word recognition phase in the model just described. However, when making plans to go swimming later in the afternoon, my friend from New Zealand wrote in an email, "*I need to go home and grab my togs before I can come over.*" Using this illustration for written language comprehension, it is possible to see how the transactional model explains the process by which I was able to infer the meaning of the word *togs*. First, while sitting at a computer checking email, visual input is received. This leads to the perceptual analysis phase, where this visual input is recognized as letters within one's language. Most of the words in the sentence were easily recognized in the word recognition phase based on their visual representation and the word meanings were quickly accessed. The word *togs*, however, was not recognized as a meaningful word based on visual representation. Based on one's understanding of sentence processing, a part of the discourse level processing, it is clear that the word *togs* represents a noun. In addition, knowledge of the world suggests that several items are associated with swimming such as goggles, bathing suits, towels, and flip flops. However, the writer from New Zealand had borrowed goggles and towels before and was regularly seen wearing flip flops during that time of year, so those necessities could be discarded from the possible items that needed to be retrieved from her home. The meaning of the word, based on analysis at the discourse level, was inferred since one of the few things that would be needed to go swimming in a neighborhood pool but would not easily accessed from my home was her bathing suit. When the inferred meaning of *togs* was inserted back into the original sentence, it was possible to comprehend the sentence, and continue making plans for later that afternoon. By using skills that are available at the discourse level of processing it is possible to infer the meaning of words that were unknown. Similarly, when one is having a conversation and comes across an unfamiliar word, it is possible to use one's understanding of sentence structure, world knowledge, and propositional knowledge to comprehend what has been spoken.

However, not all written words are easily accessed following the visual analysis stage. The process of how unfamiliar words that are not accessed from the lexicon until a phonological representation is modeled is also explained using Kamhi and Catts' model (1998). To illustrate this concept, I will share a story about my first trip to Europe. I had taken an overnight flight from Washington, D.C. to London and had planned to take a bus for several hours to meet my best friend in the little village where she was living during her year abroad. Nervous about the whole experience and suffering from a lack of sleep, I found my way through customs and attempted to locate the bus station at the airport, reading every sign that was available to me. Many of the signs in and around the airport displayed several different languages in order to assist travelers. I finally found my way to the bus station and was trying to find the line to buy the bus ticket for Norwich, but could only find signs that read, "*Queue*," which was an unfamiliar word to me at that time. I was able to perceive the visual input through visual analysis and I recognized that the print I was reading consisted of letters that are used in English, although I was not certain that *queue* was not a French or Spanish word. Since an immediate visual representation was not accessed in my lexicon, I tried sounding it out, thinking the word was pronounced as either kway-way or kwee-wee, but again, these phonological representations were not meaningful to me. Not wanting to take a bus to Queue and determined to find my way to Norwich, I looked around the bus station for someone who could help me. When I found someone who was able to help me, they responded, "You need to be in that queue over there" and pointed to the line I had almost entered. Given the context in which the word was used and my understanding of sentence structure, I chuckled to myself that I was unable to recognize the word *queue* despite having heard it used in conversation previously, and quickly went to the line to buy my bus ticket. Through discourse level processes such as world knowledge and sentence context when the word queue was spoken in a sentence, I was able to infer that that word queue was pronounced as /kju/ which was a word I had heard before but never had encountered visually. Once I had the visual input paired with the phonological representation, the word meaning was accessed, but only using both bottom-up and top-down processing according to Kamhi and Catts' model (1998).

On one level, the model set forth by Kamhi and Catts (1998) demonstrates how decoding skills and text processing work together when reading. The ability to move easily through these processes is critical to reading comprehension. Deficits in either decoding or language skills can lead to breakdowns in reading comprehension. On a broader level, the model describes the interactive relationship between oral and written language comprehension and the processes involved in each.

The Role of Phonological and Phonemic Awareness in Kamhi and Catts' Model

While not explicitly addressed, PPA is a crucial component of the transactional model of written and oral language both at the level of perceptual analysis and word recognition as depicted in Figure 1. At the earliest levels of perceptual analysis, PPA allows a listener to perceive a word and its component sounds. At the word recognition level, PPA has many roles in determining the success a reader or listener achieves in recognizing words. As noted by Catts and Hogan (2003), children who are having difficulty learning to read often have problems recognizing words, particularly with the phase of word recognition requiring decoding and the conscious application of PPA. Difficulty with decoding words limits the number of words that can be recognized immediately based on their visual representation (Ehri, 1992), and a limited number of words that can be recognized via visual representation increases demands on decoding. Poor PPA is a double-edged sword in word recognition. When a reader encounters a word that is not immediately recognized via visual representation, an attempt is made to decode the word through its phonological representation; this process requires a number of PPA-related skills. One of the skills a reader must have is letter-sound knowledge, which means the reader must know the sounds that are associated with the letters encountered in a word. In addition, the reader must have an understanding of how phonemes combine in spoken words and the skill to apply that knowledge when reading words. In order to be successful at decoding and progress in developing reading skills such that word recognition occurs as soon as a word is visualized, letter-sound knowledge and phoneme manipulation are critical.

The contribution of these PPA-related language processes in literacy development is clear. Separate from these areas of language development, but equally important to oral communication is the development of speech production skills. In Kamhi and Catts' model, speech serves as a primary form of auditory input that is received by the listener. While the reference in the model is to the receptive understanding of speech, when an individual has disordered speech, the subsequent breakdown in the communication process can influence the development of these important receptive language skills. In fact, Fey et al. (1994) indicated nearly 80% of children with delays in grammatical development also have phonological impairments. As speech input is a beginning point in Kamhi and Catts' model, understanding the relationship between receptive understanding of speech and expressive use of speech, as well as phonological awareness skills, broader oral language skills, and written language development is critical if we are to be successful in improving our understanding of literacy acquisition in children with clefts.

Phonological Awareness and Speech Disorders

Speech consists of the sounds created when airflow and energy is expelled through the oral and nasal cavities and is shaped by the lips, tongue, jaw, and velum. Speech can be impaired for many reasons. For some children there is a physical basis for their speech impairment (i.e. cleft palate, hearing impairment, and cerebral palsy), while many other children with speech impairments do not have such overt causes (Bird et al., 1995). For the remaining children without structural or motor-based limitations inhibiting speech development, a variety of patterns of speech impairments are often noted. Children may exhibit developmental articulation errors, in which immature speech patterns are present well beyond ages when their peers have outgrown these patterns. Other children may present with phonological errors, in which they adopt patterns in their speech that are not typical of any developmental pattern. Given the difficulty some children have with the development of correct representation of speech sounds, researchers have examined the phonological awareness skills of children with speech impairments and the impact these impairments have on literacy skill development.

Children with expressive phonological impairments tend to have poorer PPA skills than their typically developing peers. In one group of children ranging in age from 5;0 to 7;4, children with the most severe speech impairments at the onset of reading instruction were found to have greater difficulty in learning to read and spell than the children with mild speech impairments (Bird et al., 1995). It was the severity of the speech impairment rather than the presence of a language disorder that influenced these results, and the deficit in reading and spelling appeared to remain even after the speech impairment improved (Bird et al., 1995). Similar results were found in another study (Larrivee & Catts, 1999) that compared two groups of children, one with moderate to severe expressive phonological disorders and the other without such difficulties. The findings indicated that children who were at greatest risk for reading impairment had the most significant deficits in terms of phonological awareness, language skills, and speech productions. Additionally, those children with speech disorders with good reading outcomes had stronger PPA skills than the children with speech disorders who had poor reading outcomes (Larrivee & Catts, 1999). The results of these studies demonstrate the relationship between the severity of speech impairment and development of PPA skills.

In order to further understand the early reading skills of children with speech and language disorders, researchers have compared these children to groups of children with typically developing reading skills and those known to be at risk for reading difficulties. Carroll and Snowling (2004) compared three groups of children ranging in age from 4- to 6-years-old: (a) the first group had a history of speech and language disorders, (b) the second group consisted of children with a family history of dyslexia, and (c) the third group consisted of typically developing children. The children's language, PPA, and literacy skills were assessed. Similar patterns were noted between the children with a positive family history of dyslexia and children with speech impairment, as they had poor performance on most measures except vocabulary skills. The authors state that the results of their study "confirm the view that children with expressive speech impairments in the absence of language problems are at high risk of reading difficulties and are on a continuum with children at family risk of dyslexia" (Carroll & Snowling, 2004, p. 637).

Understanding the nature of the speech impairment is important when investigating the relationship between speech production and PPA skills. Leitao and Fletcher (2004) followed literacy outcomes in children with speech impairment from their first year in school to the end of elementary school. The children in the study were divided into two groups: one group with speech errors that were developmental in nature, and the other group exhibiting speech impairments that were not consistent with developmental patterns. All of the children completed a battery of reading, spelling, and PPA skills. Children with non-developmental speech errors showed significant deficits in measures of PPA and reading comprehension when compared to the group of children with developmental speech errors. Similar trends were noted in reading accuracy and spelling, although these differences did not reach statistical significance. These results demonstrate the importance of distinguishing between developmental and non-developmental errors in the development of PPA skills and suggest that early identification of the nature of the speech disorder can lead to more effective planning of early intervention targeted particularly at children with non-developmental errors (Leitao & Fletcher, 2004). This distinction has particular importance for children with clefts who exhibit one or both of these types of speech errors.

Sutherland and Gillon (2005) investigated PPA in children with speech impairment, comparing the performance of nine children with severe speech impairments to seventeen children with typically developing speech production skills. The investigation focused specifically on measures of phonological representation including receptive-based tasks (such as computer tasks requiring participants to judge if a pre-recorded production of the target word was said correctly or incorrectly), nonword learning tasks, and receptive gating tasks (these tasks present the initial portion of the word at varying segment lengths in terms of milliseconds and the child is required to complete and identify the word from a group of four pictured items). Production tasks, including real word and nonword repetition, were also administered. Finally, a formal test of PPA skills was administered. Results indicated that children with speech impairment had increased difficulty with receptive tasks and nonword learning tasks compared to their peers with typically developing speech. Because the relationship between speech impairments and decreased PPA skills has been well established, researchers have focused on outcome studies investigating PPA training in this population.

Phonological Awareness Interventions

Numerous studies have shown positive effects on reading skills following PPA interventions during kindergarten (Kozminsky & Kozminsky, 1995; Schneider, Kuspert, Roth, & Vise, 1997; Brennan & Ireson, 1997) and first grade (Hurford et. al, 1994).

However, the impact of PPA intervention specifically relating to children with speech impairments is of particular interest, as positive effects on both speech production and PPA skills have been demonstrated in the existing research.

For example, Gillon (2000) investigated the impact of an integrated phonological awareness intervention program for 5- to 7-year-old children with speech impairments who also demonstrated reading difficulty. Using traditional articulation therapy or minimal therapy as two separate comparison interventions, children in the experimental group who received the integrated therapy that included phonological awareness training made significant gains in measures of phonological awareness and reading development as measured by decoding skills. The children in the experimental group also demonstrated gains in their articulation skills, although they were not significantly different from the other groups. In fact, the children in the experimental group performed at levels that were similar to typically developing children at post-test assessments of PPA skills (Gillon, 2000). Additionally, in a follow up study conducted approximately one year after the post-intervention assessment, Gillon (2002) reported that the children in the experimental group demonstrated sustained effects of their intervention, as demonstrated by the fact that many of these children were reading at or above their age-levels on measures of word recognition, and that their phoneme-grapheme connections had remained strong when these skills were reassessed. Conversely, the children in the comparison groups that did not receive PPA intervention continued to be classified as "poor readers" (Gillon, 2002). The results of these studies show that given the risk of decreased PPA skills in children with speech disorders, therapy that addresses both the articulation as well as PPA skills has the potential to improve both sets of skills and have a lasting impact on reading development.

Gillon (2005) also followed the PPA development of a group of children with moderate to severe speech impairment who were identified when they were 3- to 4-years old. Children with speech impairments were compared to a group of peers with typically developing speech over three points in time. Intervention for the children with speech impairment lasted anywhere from 16 to 34, 45-minute sessions, with the average number of sessions being 25.5. Therapy sessions blended speech and PPA tasks into the same activities. Speech intervention followed a cycles approach with techniques including auditory bombardment and drill play activities. Phonological awareness intervention activities included phoneme detection, phoneme categorization, initial phoneme matching, and phoneme isolation. Older children in the intervention were exposed to segmentation and blending tasks, as well as letter-name and letter-sound tasks. Gillon concluded that this type of intervention not only improved speech intelligibility while stimulating phoneme awareness in children as young as three to four years old, but this type of intervention was helpful with early reading and spelling performance when the children were 6 years old (Gillon, 2005).

In an earlier study, Hesketh, Adams, Nightingale and Hall (2000) reported findings that appear to conflict with those cited by Gillon (2000; 2002; 2005). Hesketh et al. compared outcomes in 3 ½ to 5-year old children with developmental phonological disorders following two different intervention types: the first intervention was a metaphonological therapy approach and the second was an articulation-based therapy. The children in the articulation-based therapy treatment group participated in activities focusing on production of phonemes or phoneme classes in consonant vowel or vowel-consonant combinations, moving to word and sentence productions. The children in the metaphonological treatment first practiced awareness tasks such as rhyming, syllable segmentation, alliteration, blending, and segmenting for the initial four weeks of the treatment. During the subsequent four weeks, participants engaged in these activities using the phonemes that had been the target of the phonological intervention. During the last two weeks of the intervention, therapy focused on production of contrasting sounds and minimal pairs, although there still was no direct correction of speech (Hesketh et al., 2000). Results indicated the children receiving the direct articulation therapy made the most progress on speech outcome measures. However, there were no significant differences between the articulation and metaphonological group on measures of PPA skills when assessed three months following this intervention. The authors also note that the children in the articulation therapy condition made gains in metaphonological skills even without explicit metaphonological intervention, as traditional articulation therapy inherently focuses on speech sound awareness.

Findings presented by Hesketh et al. (2000) appear to be in conflict with Gillon (2000; 2002; 2005). While Gillon consistently demonstrated the benefits of PPA training, Hesketh et. al found that speech production and PPA skills were not significantly different following two different interventions that focused either on articulation or PPA skills. The reasons for the differences in findings become more apparent when the methods are carefully examined. First, across studies, Gillon (2000; 2002; 2005) describes an intervention that features phonological awareness tasks that were integrated into therapy that also actively addressed speech production. In comparison, Hesketh et al. contrasted an articulation-based approach with a PPA skill driven approach that did not address speech production. In other words, Hesketh et al. did not investigate any integrated approaches. Therefore, the types of PPA interventions reported by Gillon and Hesketh et al. were different. Secondly, it is important to consider the ages of the children who participated in the two studies. The children in Hesketh et al ranged in age between 3 ½ and 5 years old while the participants in Gillon's studies were between 5- and 7-years-old (Gillon 2000, 2002). The difference in ages is an important distinction as children who are slightly older are more likely to respond to PPA training. Gillon (2005) provided an integrated PPA and speech production treatment to children with identified speech deficits beginning when they were 3- and 4-years-old and continuing until they were 5-years-old, but the impact of these interventions was measured when the children were 6-years-old. Even when Gillon provided an intervention targeting children the same age as Hesketh et al., her assessment occurred when children were older, once again creating an age difference between the two subjects. Given the differences in study design and the age range of children, it is possible to understand how these studies present such different findings.

While there is clear evidence to support the incorporation of PPA training in therapy aimed at improving speech production, most studies investigating PPA in children with speech disorders specifically exclude children with structural anomalies such as cleft palate. When children with clefts present with speech disorders, the errors may or may not be related to the structural defect. However, when the error is due to the structural defect, many children adopt systematic patterns to compensate for their inability to produce the target phoneme appropriately. Surgical intervention aimed at repairing the cleft defect can limit the development of compensatory patterns, but there is a need to balance the importance of cosmetic appearance with function for breathing, speech, and feeding skills. Every effort is made to ensure proper velopharyngeal functioning prior to a child entering school; however, children with clefts often have differences in jaw positioning and dentition that potentially impact speech well into their school age years and in some cases, even into young adulthood.

Given the unique factors influencing the speech of children with clefts, it is important to explore this issue further.

Speech Production in Children with Clefts

A cleft is "an abnormal opening or a fissure in an anatomical structure that is normally closed" (Kummer, 2001, p.52). In the embryological period, a cleft is the result of a failure of the palatal shelves to fuse within the first twelve weeks of life. Clefts of the lip and palate vary in severity from mild involvement, which may go undetected by a layperson, to more severe clefts that are wider, longer, and involve more tissue, thus affecting cosmetic appearance and function of the mechanism for speech and feeding purposes. However, just because a cleft looks relatively mild, the underlying musculature may still be malformed, significantly impacting the function of the mechanism.

A cleft lip typically involves the upper lip and can be unilateral or bilateral, although occasionally a midline cleft does occur. It is typically closed surgically around three months of age, although the timing of surgery varies according to the health of the baby, the size of the cleft, and the recommendations of the cleft palate team treating the child. A cleft lip may present in its mildest form, an incomplete cleft lip, as a small notch in the lip with no related impairment to function. Conversely, in the case of a complete cleft lip, the cleft extends through the muscles of the lip into the nostril and potentially involves the primary palate, which includes the alveolus and the portion of the palate anterior to the incisive foramen (Kummer, 2001). A complete cleft lip might result in some initial difficulty with infant feeding, but, once repaired, usually has a minimal effect on speech development as the only consonants that require lip closure are /p/, /b/, /m/, and /w/. Although rare in the United

States, a cleft lip unrepaired can impact speech skills as well as dentition, making the production of these phonemes even more difficult.

A child with a cleft lip may or may not also have a cleft palate. A cleft palate creates an opening between the oral cavity and nasal cavity. Like a cleft lip, a cleft palate, presents with different degrees of severity; it can occur in isolation or in addition to a cleft lip. An isolated cleft of the palate can be associated with a syndrome, which "involves a pattern of multiple anomalies that regularly occur together and…have a common known or suspected cause" (Kummer, 2001, p. 505). In its mildest form, a cleft palate presents as a bifid uvula, while the most severe form presents with a cleft extending from the incisive foramen posteriorly through the hard and soft palates. A cleft palate is typically repaired surgically around one year of age, again depending on factors such as the general health of the child, the size of the cleft (O'Gara & Logemann, 1988), and the recommendations of the treating team.

A child with a cleft palate is unable to produce consonants that require oral pressure build up, as the child lacks the oral and velopharyngeal structures to separate the oral and nasal cavities. Even after the initial palatal repair surgery, children with cleft palate may require secondary surgeries in order to provide an adequate mechanism for speech. An individual with velopharyngeal inadequacy or insufficiency may develop obligatory errors (including hypernasality, nasal air emission, and weak pressure consonants), as well as optional/learned errors that include compensatory misarticulations and phoneme specific nasal air emission (Trost-Cardamone & Witzel, 2000).

Consonants present particular difficulty for children with cleft palate, even after the initial repair. A consonant is a phoneme that involves constriction of the vocal tract (Small, 2005). In English, there are sixteen oral pressure consonants: /p/, /b/, /t/, /d/, /k/, /g/, /f/, /v/, /s/, /z/, “sh”, “zh”, “ch”, “j”, voiced “th”, and voiceless “th”. To produce a consonant, airflow travels up from the lungs and passes through the vocal folds, which either vibrate or not in order to produce voicing. The stream of air then enters the pharyngeal cavity. For oral pressure consonants, the velum must elevate in order to close off the oral and nasal cavities. The airflow and resonating energy is then shaped at various places within the oral cavity and a phoneme is produced. Consonants vary in terms of place, manner, and voicing. Place refers to where the phoneme is formed, manner refers to how the phoneme is formed, and voicing refers to vocal fold vibration (Shriberg & Kent, 2003). Precise movements of the articulators, including the tongue, jaw, lips and palate result in the production of phonemes that are recognized by speakers of a shared language. Given this, it is easy to understand why consonants pose a particular challenge to children with cleft palate.

Children with repaired cleft palate often exhibit similar consonant error patterns. Obligatory errors, or passive errors, occur secondary to a structural defect such as velopharyngeal dysfunction or a palatal fistula (Peterson-Falzone, Trost-Cardamone, Karnell, & Hardin-Jones, 2006). Features such as nasalization of oral pressure consonants (i.e. /b/ produced as /m/; /d/ produced as /n/) typically fall into this category. Nasalization of these sounds results in the individual using the correct place of constriction within the oral cavity, but due to faulty velopharyngeal valving, a nasalized phoneme is produced. Another obligatory error is hypernasal resonance, which is excessive energy enhancement in the nasal cavity during the production of vowels and vocalic consonants. Additionally, weak pressure consonants result when the speaker has reduced build up of oral pressure during the production of oral pressure consonants (Peterson-Falzone et al., 2006). Finally, nasal air emission results when airflow escapes from the nose during an attempt at an oral pressure consonant. Each of these obligatory errors influences speech production and occur without conscious effort by the speaker.

Compensatory errors are a separate class of errors often observed in the speech patterns of children with clefts. Compensatory errors are those errors produced by a child actively attempting to compensate for the faulty velopharyngeal mechanism, and they occur frequently in speakers who are aware of their difficulty generating oral airflow for speech purposes. A child who uses compensatory patterns backs up the place of production to a location in the vocal tract that is before any faulty mechanism. In the case of an individual with a cleft palate and velopharyngeal dysfunction, oral consonants may be moved to the glottis as a substitution for a sound that requires pressure build up within the oral cavity, resulting in a sound that is perceptually similar to the target phoneme by many listeners (Peterson-Falzone et al., 2006). Other examples of compensatory errors in young children with clefts include pharyngeal fricatives, pharyngeal stops, coarticulations, and active nasal fricatives (Harding & Grunwell, 1998). Pharyngeal fricatives and pharyngeal stops are produced with contact between the tongue base and the posterior pharyngeal wall; individuals use these sounds, much like glottal stops, when they are unable to achieve pressure build up at the appropriate place of articulation. Coarticulations typically employ a glottal stop (although pharyngeal stops may also be used) in addition to the appropriate place of production for the target phoneme, but secondary to inability generating oral pressure, the glottal stop is used in order to have some level of plosiveness. Active nasal fricatives refer to the process in which an individual releases airflow voluntarily through the nose while closing the oral cavity; this pattern is often used as a substitution for sibilant sounds (Harding &

Grunwell, 1998; Peterson-Falzone et al., 2006). Use of these compensatory phonemes require the speaker to use alternate placement of their articulators in an attempt to produce distinctive sound productions when velopharyngeal function does not allow for adequate production of the target sounds. Given the manner in which children with clefts compensate for and otherwise develop atypical speech production, it is prudent to understand how and if these patterns have an impact on the development of PPA and written language.

Advice regarding the treatment of obligatory and compensatory errors in children with repaired clefts is readily available. There are a number of textbooks that either include treatment techniques as part of the overview of speech disorders relating to cleft palate (Bzoch, 1997; Kummer, 2001; Peterson-Falzone, Hardin-Jones, & Karnell, 2001), or devote the entire content of the book to therapy techniques that are useful with this population (Golding-Kushner, 2001; Peterson-Falzone et al., 2006). The authors of these texts rely heavily upon evidence from their own clinical experience as the foundation for their recommendations regarding treatment. As a result, the treatments described in these books reflect the opinions of the authors regarding techniques that are effective and those that are of little benefit to this population.

In *Therapy Techniques for Cleft Palate Speech and Related Disorders*, Golding-Kushner (2001) provided nearly 200 pages of information relating to the treatment of speech and resonance disorders in individuals with cleft palate. The author included a chapter entitled, “Evaluation and Therapy Techniques to Avoid”, where she discussed strategies that she has not found to be beneficial. Regarding the use of techniques such as sign language and oral motor therapy, Golding-Kushner stated:

> There have been no rigorous scientific investigations that would suggest their advantage. Stated more strongly, these techniques have a long history of failure. On the other hand, the direct articulation therapy techniques described throughout this volume have been applied successfully to thousands of patients with a high degree of success. (p. 133)

While this statement may be true given the author's clinical experience, the author does not support her recommendations for direct articulation therapy with rigorous scientific investigations.

Similar recommendations are found in *The Clinician's Guide to Treating Cleft Palate Speech* by Peterson-Falzone et al., (2006). These authors also caution against the use of oral motor exercises, reminding the reader that children with cleft palate typically do not have difficulty with velopharyngeal strength as much as they have palates that are of inadequate length for velopharyngeal closure (Peterson-Falzone, Trost-Cardamone, Karnell, & Hardin-Jones, 2006). Instead, the authors advocate teaching the child about the structures involved in speech, expanding the child's phonemic inventory, and teaching the child to be aware of the difference between oral and nasal sound productions.

Direct articulation therapy is frequently the approach advocated by many speech-language pathologists with years of experience in the treatment of speech disorders in children with clefts (Golding-Kushner, 2001; Peterson-Falzone et al., 2006). For example, Trost-Cardamone and Witzel (2000) recommended traditional articulation methods such as establishing a "place map" and producing the target at the syllable level consistently, before moving into word-, phrase- and sentence- levels of production. Hobbs, Moore, & Salomonson (2002) advocated for speech therapy to occur for one hour twice a week focusing on multiple productions of the targets. While clinical experience has likely lead to these recommendations, the scientific evidence for these claims is lacking.

In a review of the literature examining evidence-based treatment approaches for speech development in individuals with clefts, only four intervention studies were identified for preschool and school age children, using two different approaches: phonological approach and intensive articulation therapy. Hodson, Chin, Redmond, & Simpson (1983) and Pamplona, Ysunza & Espinosa (1999), presented studies that demonstrated the efficacy of phonological approaches to speech intervention for children with clefts. Albery and Enderby (1984) and Van Demark and Hardin (1986) compared the efficacy of intensive speech therapy (such as speech camps) using traditional articulation therapy approaches to therapy programs that were provided using a weekly therapy model. Following intervention, Albery and Enderby (1984) reported the number of errors in the experimental group that received intensive speech therapy was significantly lower than that of the control group who participated in community-based services. Van Demark and Hardin (1986) used a similar intensive treatment model, although they did not find significant differences in outcome following the intervention.

Regardless of the treatment model used to address speech production, the overall goal of speech therapy when working with children with clefts is to improve articulatory movements while eliminating compensatory patterns. Therapy is usually recommended when there is no need for surgical management of velopharyngeal function (Kummer, 2001) or when surgery is not indicated (i.e. the child is medically fragile) yet articulatory placement errors exist. While normalized speech production prior to entering school is commonly advocated, it is not always realized. As a result, children with clefts may have speech production errors that result from structural differences that prevent the production of oral consonants, or they may have errors that are the result of learned behaviors that remain following surgical correction. While preschool and early school-age children with clefts who present with persistent speech errors are likely to be involved in speech therapy focusing on the remediation of these errors, the impact of these errors on PPA skills remains unknown as does the impact of a great number of intervention approaches suggested in clinical textbooks.

Phonological Awareness and Oral Language

In addition to speech production, language skills are also a critical component of communication, speech, and written language. While speech consists of the sounds in a spoken language, language "is a code whereby ideas about the world are expressed through a conventional system of arbitrary signals for communication" (Lahey, 1988, p. 2). Language is often conceptualized as having three dimensions: content, form, and use. Content refers to the meaning behind what is being discussed. Form refers to the rules that govern the language, including phonology, morphology and syntax. Use refers to how language is used to achieve goals in communicating (Lahey, 1988). These linguistic concepts of content, form, and use have their place in Kamhi and Catts' model of Spoken and Written Language Comprehension (1998). Linguistic competency relies on an individual's ability to master each of these skill sets. If there is a breakdown in one's command of the content, form, or use of the language, the intended message may not be interpreted correctly.

Individuals with reading difficulties commonly have deficits in oral language (Catts, 1993; Catts, 1996; Catts, Fey, Zhang, & Tomblin, 1999; Catts & Hogan, 2003; Snyder & Downey, 1997) across one or more dimensions. While there are many aspects to explore in the relationship between oral language disorders and reading, the specific relationship between language development and PPA skills is of particular interest when investigating literacy skills of children with clefts, as this relationship has not yet been explored.

Phonological Awareness and Language Disorders

The relationship between oral language and PPA has been demonstrated in a number of ways. At the primary grade levels, PPA has been linked to oral language through a longitudinal investigation of the relationship between oral language and PPA (Cooper, Roth, Speece, & Schatschneider, 2002). The 52 children in this study were assessed during kindergarten and again in second grade on measures of family background, oral language skills and PPA skills. Regression analysis revealing that general oral language was responsible for unique variance in PPA ability indicates that oral language skills contribute to the development of PPA skills from kindergarten through second grade (Cooper et al., 2002). Similar findings resulted when Rvachew (2006) investigated factors that influence PPA skills in a group of 47 kindergarteners with speech production disorders. Testing was administered during spring or summer before the child entered kindergarten and again during the spring and summer of the child's kindergarten year. Assessments included measures of speech perception, vocabulary, articulation and PPA skills including rime matching, onset matching, and onset segmentation matching. Correlational analyses revealed significant positive relationships between speech perception skills and receptive vocabulary on PPA at the end of the kindergarten year (Rvachew, 2006). These studies demonstrate the important links between language skills and PPA skill development.

Using a broader range of reading outcomes measured across kindergarten, first, and second grade, Catts (1993) investigated the relationship between speech and language impairments and reading disabilities. Children in the study were first tested in kindergarten using two tasks of PPA (syllable or phoneme deletion and sound blending tasks) and rapid automatized naming; they also received a battery of standardized receptive and expressive language assessments targeting vocabulary, syntax, morphology, and speech production skills. The children were then tested again in first and second grades using various standardized reading assessments. The impact of speech, language, and PPA skills on reading depended on the reading outcome of interest. For example, Catts reported that word recognition skills at the single word level and in context were strongly influenced by PPA skills and rapid automatized naming; however, language skills alone were more important to measures of reading comprehension.

This relationship between PPA, oral language, speech and reading skills was also found in a study of adolescents who had a history of speech and language impairment as preschoolers (Stothard, Snowling, Bishop, Chipchase, & Kaplan, 1998). According to the results of the investigation, those adolescents whose language deficits had not resolved by 5 ½ years of age performed significantly less well on PPA and literacy assessments, indicating that early speech and language deficits may have an ongoing impact on educational outcomes. Conversely, those with language deficits that had resolved by 5 ½ years of age had better outcomes. Taken collectively, the results of these studies suggest that children with language deficits are indeed at risk for difficulty with reading, whether it is due to a core PPA deficit or because their language skills do not support their comprehension of the written text.

Based on the model presented by Kamhi and Catts (1998), it is possible to see how both PPA and language skills are critical to written language comprehension, as one uses a top-down approach, and the other uses a bottom-up approach. When one encounters an unfamiliar word, PPA skills can be used to derive meaning when decoding by accessing the word meaning through a phonological representation. However, one can also use discourse level processes such as understanding of sentence structures, morphological markers, and the overall context in which the word is being used in order to access word meaning. Language skills, as well as PPA skills, are critical to early reading development, as comprehension of oral language is needed in order to derive meaning from the word that is being decoded.

Language Development in Children with Clefts

Given the importance of oral language skills in literacy development, it is necessary to explore these skills in children with clefts. Compared to the body of literature that exists regarding speech development in this population, there has been relatively little emphasis on language development in children with clefts. Referring back to Kamhi and Catts' model (1998), one can see that understanding at the single word level as well as in the sentence/discourse level is bi-directional and thus critical in comprehension of both oral and written language. For that reason, it is important to explore what is known about oral language skills in children with clefts.

Delays in early language development for children with clefts are not uncommon. Given the disruption in speech development due to abnormal anatomical structures as well as the high number of middle ear infections they face, it should not be surprising that children with cleft often experience delays in language development. In a recent review of the literature, Sharp, Dailey, and Moon (2003) suggest that reports of deficits in expressive language skills are quite possibly the result of the difficulty children with cleft have with speech production. They cite an example of the inability to produce the phoneme /s/, resulting in the inability to use plural "s" markers as well as possessive "s". They additionally cite the frequency of ear infections as leading to periods of decreased auditory perception as another factor impacting language development. While speech impairment and decreased hearing acuity are likely to contribute to the differences in language skills, the extent to which they are responsible for the differences is unclear.

Language Skills in Infants and Toddlers with Clefts

The nine studies identified that examined the language skills of children with clefts between the ages of six and thirty-six months suggest that the language skills of children with clefts tend to fall below average but within normal limits. Long and Dalston (1982) compared the gestural communication of ten children with cleft lip and palate to ten children without clefts who were all between 12 and 13 months of age. In this study, the researchers sampled the interactions between mothers and their children, examining variables such as pointing, showing, giving, reaching, refusing, and showing off. Long and Dalston concluded that there were no significant differences across these variables between the two groups of children. This is an important finding, as gestural communication is an important prelinguistic form of communication. The findings also suggest that interactions between infants and their mothers are not having a negative impact on the language development of young children with clefts.

Seven of the eight remaining studies in the infant/toddler age bracket extend these findings to support the understanding that children with clefts often perform at levels that are lower than their peers although not necessarily outside normal limits. Jocelyn, Penko, and Rode (1996) reported their findings regarding the receptive and expressive language skills of children with cleft lip and palate at one and two years of age. The scores of the children with cleft lip and palate were significantly different from the comparison children on measures of language development. However, differences between the two groups identified in this study were not demonstrated in spontaneous language measures such as mean length of utterance (MLU). Jocelyn et al. also reported scores from general measures of child development noting that the children with cleft lip and palate scored significantly lower than the comparison group. These findings were confirmed by Broen et al. (1998) who reported that children with clefts performed within normal limits on a similar battery of language and developmental assessments while still falling below the comparison groups on each of the measures. More recently, Speltz et al. (2000) used the same measures of language development and noted that there was no difference in performance on the language measures at 12 months of age, but the children with clefts did score lower than their comparison group on measures of expressive language at 24-months of age. Fox, Lynch and Brookshire (1978) also reported mild delays of one-to-three months across the three language and developmental screening tests administered to children ranging in age from two to 33-months. Using parent report, Neiman and Savage (1997) collected data on a variety of developmental skills, noting that children with clefts in their study exhibited delays or at risk language skill levels at two points in time—5 months and then again at 36 months. These studies consistently support the conclusion that while children with clefts have spoken language skills that are within the average range, there is a consistent trend that demonstrates their skills are below those of their peers.

Morris and Ozanne (2003) asked a slightly different question than these other studies that were simply attempting to determine if the children with clefts performed differently than the children without clefts. Rather, they compared the performance of children with clefts who were identified as having delayed expressive language skills at 2 years of age to peers with clefts that did not have expressive language delays at 2 years of age. At 3 years of age, these two groups completed a test battery examining several speech and language skills, with significant differences noted in MLU as well as in speech production skills. This led Morris and Ozanne to conclude that there is a subgroup of children with clefts who are at risk for ongoing deficits in speech and language development. Consistent with the findings of reduced mean length of utterance, Chapman, Hardin-Jones, and Halter (2003) indicated that in spontaneous speech samples taken with children and their caregivers, children with cleft lip and palate used fewer words than their non-cleft peers. Taken as a whole, the results of these studies suggest that children with clefts do not appear to reach the same levels of oral language achievement as their matched peers.

Only one study has failed to find any differences in the oral language skills of children with clefts (Starr, Chinsky, Canter, & Meier, 1977). When the performance of the children with clefts in this study were compared to the normative data for a standardized assessment of development, no significant differences were observed on the mental or motor functioning domains for children who were followed from 6-months of age to 2 years of age. However, raw data included in this study support the notion that there is a trend toward lower performance, as the group means consistently fell below the average scores listed as normative data for each age range.

Despite the findings that suggest a trend toward at risk or delayed language skills in infancy and toddlerhood for children with clefts, it is important to look at methodological differences between these studies. First, the size of these studies varied greatly. The smallest study included only nine children in the experimental group (Morris & Ozanne, 2003), while the largest study included 186 children with clefts (Neiman & Savage, 1997). Secondly, while sample size plays an important role in the strength of conclusions that can be drawn from the study, the use of comparison groups is also important. For example, even though Morris and Ozanne had only nine children with clefts in their experimental group, their comparison group consisted of children with clefts as well. By using children with clefts as the comparison group, variables such as the presence of a cleft contributing to an overall delay in early toddlerhood or matching for risk factors such as middle ear infections are controlled. Thus, the conclusion of a subgroup of children with clefts with language delays that continue into early preschool is a fairly strong conclusion. In contrast, Neiman and Savage employed a much larger sample size, but the design was cross-sectional and compared results to normative data rather than longitudinal, as was the case in some of the smaller studies. The majority of these studies had comparison groups that were matched on factors such as age, gender, race, and some level of socioeconomic status (SES) such as mother's education level or an unspecified measure of SES (Chapman et al., 2003; Broen et al., 1998; Long & Dalston, 1982; Jocelyn et al., 1996; Speltz et al., 2000). Fox, Lynch, and Brookshire (1978) only provided information that the children in the comparison group came from a local hospital. Only two studies compared their experimental subjects to developmental norms (Neiman & Savage, 1997; Starr et al., 1977). Given the subtle differences in language skills that are often reported in children with clefts, ensuring that the comparison group is well-matched to the experimental group as well as using sufficient sample size is critical.

An additional factor to consider when evaluating the strength of the conclusions drawn from these studies is the inclusion of multiple cleft types within the experimental group. As mentioned previously, children with clefts constitute a very diverse group. Some studies have deliberately excluded particular cleft types in order to attempt to decrease some of this diversity. Only two studies reviewed here included children with cleft lip only (Neiman & Savage, 1997; Starr et al., 1977); these children are rarely included in current studies given the low rates of speech or language difficulties and it might reasonably be speculated that their inclusion skewed the data to reflect less of a delay. Additionally, Chapman et al. (2003) and Jocelyn et al. (1996) excluded children with cleft palate only. This is likely due to the notion that children with cleft palate only also have a higher risk of other associated syndromes. Interestingly, only one of the studies reviewed here reported data that supported the traditionally held view that individuals with cleft palate only are at a higher risk than other cleft types for language delays (Neiman & Savage, 1997); the remaining studies that separated the children into groups based on cleft type did not find significant differences between children with cleft palate only and cleft lip and palate (Starr et al., 1977; Speltz et al., 2000; Broen et al., 1998). Fox et al. (1978) presented the only study that included children with cleft palate only but did not report the results separately. Given the age of the study, it is possible that cleft palate only was not as commonly associated with syndromes at that time. Despite the fact that these studies did not find cleft type to have an impact on language delay, the commonly held opinion among professionals working with children with clefts is that children with cleft palate only have a higher risk for language delay.

In summary, the literature suggests that infants and toddlers with clefts are at risk for decreased language skills when compared to their peers. It is important to note that these language skills, while typically lower than same age peers without clefts, do fall within normal limits, albeit the low end of the limits. Studies that treat all children with clefts as one large homogenous group rather than presenting data according to cleft type or in relation to hearing status make it difficult to determine the cause of any delay, if detected at all. Additionally, while a variety of measures were used to assess language skills, the finding of decreased language skills remained consistent.

Language Skills in Preschool and School Age Children with Clefts

Seven studies that examined oral language skills in preschool and school age children with clefts were identified, with all seven noting some difference in language form, content or use. Two of the studies found overall delays in language skills in general (Philips & Harrison, 1969; Smith & McWilliams, 1968), while two additional studies reported delays in both receptive and expressive vocabulary (Nation, 1970; Brennan & Cullinan, 1974). In a study that compared the language skills of children with clefts ranging in age from 18- to 72-months, Philips and Harrison (1969) found that children with clefts performed at levels that were significantly different from children their age without clefts on measures of receptive and expressive language, with most significant delays noted for children below three years of age. Smith and McWilliams (1968) also noted an overall delay in language skills in children ages 3 years to 8 years 11 months, but results of their study contradicted the finding of the previous study, in that the Smith and McWilliams study noted that language deficits in their sample seemed to increase as the child matured. Nation's (1970) study only examined vocabulary skills in preschool children with clefts, noting that in general, children with clefts had delayed vocabulary skills when compared to their siblings as well as in comparison to the normative data available on the assessments used in the study. Nation's findings, while noting overall delays in both receptive and expressive vocabulary, indicated more significant deficits in terms of expressive vocabulary. Similarly, Brennan and Cullinan reported decreased receptive and expressive vocabulary skills in children with clefts when compared to a comparison group that was matched for gender and age.

Several studies suggest that children with clefts have delays in expressive language skills only. Spriestersbach, Darley, and Morris (1958) found that while the children with clefts in their sample ages 3 ½ years to 8 ½ years had good receptive vocabulary skills as well as grammatical skills that resembled normative data, their expressive vocabulary skills were below average. Pamplona, Ysunza, Gonzalez, Ramirez, & Patino (2000) found that the presence of compensatory speech errors in their sample of 3 to 8 year olds was associated with decreased expressive language skills when compared to other children with clefts who did not have speech errors; receptive language skills were not assessed in this study. Finally, Chapman et al. (1998) examined the conversational skills of preschool and school age children with clefts compared to their peers without clefts. While there was no significant difference reported in terms of level of participation, preschool children with clefts did demonstrate a less assertive style of communication. As demonstrated by these studies, delays or differences in language development that appear to emerge in the infant and toddler period remain well into the preschool and school age years, particularly with respect to expressive language skills.

A major weakness in the collective findings of these studies is the age of the studies; five out of the seven studies described here were published between 1958 and 1974. As mentioned with the discussion on infant and toddlers, at the time when these older studies were published, less was known about the diversity of these patients based on cleft type. In fact, only the two most recent studies limited their subjects to unilateral cleft lip and palate (Pamplona et al., 2000; Chapman et al, 1998), while the other studies included individuals with a variety of cleft types. Limiting the inclusion criteria to one cleft type or reporting data according to groups based on cleft type helps to decrease the variability of the subjects and strengthens the conclusions made by the researchers.

As with the infant and toddler group, the number of experimental subjects included in these studies was highly variable, ranging from 14 children with clefts (Brennan & Cullinan, 1974) to 137 children with clefts (Philips & Harrison, 1969). Comparison groups were used in the majority of studies, although the older studies provide little information on the criteria used to match the children in the comparison groups. Nation (1970) used two comparison groups in his study: the first comparison group consisted of siblings of the experimental group, while the second comparison group was simply described as a group of "normals." As stated before, Pamplona et al. (2000) used a group of children with a history of cleft palate but without compensatory articulation errors to determine the extent to which these speech errors relating to the cleft were impacting language skills. The lack of information provided regarding the comparison groups is likely a factor of the age of most of these studies. Greater weight should be given to the findings that provided information about well-matched comparison groups.

In summary, these studies indicate that children with clefts are at risk to have deficits in language skills that persist into their preschool and school age years. While the results suggest both receptive and expressive language skills are at risk, expressive language skills in children with clefts appeared to be more consistently impacted. Given the relationship among language skills, PPA, and written language described in the theoretical framework underlying this study, it is reasonable to hypothesize that children with clefts would have difficulty with PPA and written language development in light of the language deficits they face.

One important aspect of receptive and expressive language development that was not explored in the aforementioned studies of language development of children with clefts is hearing status. Hearing status is also not addressed explicitly in the model of spoken and written language that serves as the theoretical framework for the current investigation. Yet, hearing status is clearly related to spoken and written language development and must be addressed when investigating speech and language-related skills in all children.

Investigating the hearing status of children with clefts when investigating language-related skills is particularly important given their increased risk for increased otitis media, which is related to mild conductive hearing loss and differences in language development. Referring back to Kamhi and Catts' model of spoken and written language, it is possible to see how differences in auditory perception can lead to breakdowns in comprehension of spoken language. If the auditory signal is not adequately perceived, there are implications for the auditory analysis and phonological representation phases in spoken language comprehension. Weak or inadequate phonological representations resulting from poor auditory analysis impact not only word meaning, but also have implications for comprehension of written language.

Impact of Otitis Media on Language Development

The impact of otitis media on language related skills has been the subject of considerable controversy, however, it is frequently cited as a potential cause of language differences in children with clefts (Broen et. al, 1996). Otitis media is a condition in which the middle ear space, which is normally filled with air, becomes filled with fluid. This fluid can be infected, as in the case of acute otitis media, or the fluid may not be infected, as in the case of otitis media with effusion (Roberts & Hunter, 2002). The presence of fluid in the middle ear space, besides potentially causing ear discomfort in young children, has the potential to cause a conductive hearing loss due to the fact that sound is not as easily transmitted through the middle ear to the inner ear when there is fluid, as opposed to air, filling the space. The traditionally held concern as it relates to the development of communication is that this hearing loss interferes with a child's ability to hear the discrete differences in speech and language and may therefore lead to delays in development. When this difficulty perceiving speech is coupled with the difficulties many children with clefts experience when producing speech, it is understandable that PPA and other language skills are impacted.

According to Kamhi and Catts' model of spoken and written language comprehension, a child must recognize a sound signal as a discrete unit of speech and then encode that sound with the corresponding letter (Kamhi & Catts, 1998). When a child has difficulty perceiving sounds in oral language, the process is disrupted, and it is difficult for that child to assign a letter to that sound in written language. This difficulty then disrupts the entire process of language acquisition.

Results of studies that have examined the short- and long-term impact of early and prolonged experience with otitis media on language related skills such as early speech development, morphological and syntactical development, vocabulary, language use, and attention are inconsistent in the general population. Additionally, the measures used to assess these skills vary from study to study, making the interpretation of the conflicting results challenging. Using newer theoretical models, such as the Contextual Transactional Model (Vernon-Feagans, Miccio, & Yont, 2003) helps to explain these differences across studies. The Contextual Transactional Model begins with the concept that otitis media leads to hearing loss, but also allows for the consideration of mediators and moderators that may also impact the child's ability to attend to language. This attention to language is then thought to lead to differences in speech, use of language, and the ability to sustain attention to language. Early, fluctuating hearing loss might make it difficult to attend to language, particularly in environments with a large degree of background noise. The demands of attending to language in a noisy environment might lead to periods of inattention that might in turn have negative effects on language development as well as social interaction patterns (Creps & Vernon-Feagans, 2000). The Contextual Transactional model concedes that language skills may ultimately recover, as has been demonstrated by Roberts et al. (2000) and Roberts, Burchinal, and Zeisel (2002), but the child's use of language in social interactions and ability to maintain attention in noisy situations may be negatively influenced over the long term. Given that this model allows that moderators may decrease the impact of otitis media on language related skills, strategies to modify the home, daycare, and even classroom environments can facilitate positive outcomes.

Despite the common concern that the presences of otitis media has the potential to lead to a mild hearing loss, there were few studies that examined the degree of hearing loss in young children who suffered from otitis media. Gravel and Wallace (2000) determined that children who had a low incidence of otitis media episodes were found to have hearing threshold levels within normal limits (between 0-20 dB), while 30-45% of children with a high incidence of bilateral otitis media had hearing thresholds greater than 20 dB. Hearing levels with unilateral or infrequent otitis media were shown to fluctuate, although they typically remained within normal limits. This is an important finding as it confirms that children with a history of otitis media, particularly those with bilateral otitis media, are indeed at risk for decreased levels of hearing sensitivity.

Beyond the association between otitis media and a decrease in hearing acuity levels, there is little agreement regarding the effects of this condition on developmental skills relating to language. For example, a significant history of otitis media in infants has been linked to limited phonetic inventories (Petinou, Schwartz, & Mody, 1999), while other studies have noted that as long as hearing remains within normal limits, phonetic inventories, specifically consonant inventories, reflect those of infants without a history of otitis media (Miccio, Gallagher, Grossman, Yont, & Vernon-Feagans, 2001). Otitis media has also been found to have a short-term effect on language development for young children. Differences have been noted in children with histories of otitis media in auditory perception tasks (Petinou, Schwartz, Gravel, & Raphael, 2001), receptive language skills (Paradise et al., 2000) and overall development of language skills through three years of age (Feldman et al., 2003). While the differences presented here were generally small, the children did show deficits in speech production, speech perception, receptive language, and expressive language skills. Despite the consistency of these findings, it is important to note that several of the authors reported that additional factors, such as socioeconomic status, contributed to the observed differences in speech and language skills in toddlers (Feldman et al., 2003; Paradise et al., 2000).

While otitis media episodes usually decrease in frequency and duration as children age, the potential long-term effects have been studied in relation to speech and language related skills. Episodes of otitis media are rare in children by age 7 (Teele et al., 1990). However, a significant history of early otitis media may have an impact on speech and language skills that remains well past age 7. In an extensive review of the literature, Vernon-Feagans (1999) cited only two articles that found that children with a history of otitis media had differences in speech production.

There has been much more focus, however, on the impact of otitis media on language skills. The results continue to be mixed. Several studies have found no significant differences on standardized language tests for children with a significant history of clefts. Gravel and Wallace (2002) report that while differences were not noted in terms of receptive and expressive language skills, children with histories significant for otitis media exhibited differences in their ability to attend in noisy backgrounds. Roberts et al. (2000) reported correlations between hearing loss and the presence of persistent otitis media to scores on language measures at 3- and 4-years of age; however, these results were not significant once environmental factors such as poverty and supportive home environments were considered. Additionally, in a follow-up study with the same group of children, Roberts, Burchinal, and Zeisel (2002) indicated that children with a history of otitis media in early childhood who exhibited expressive language skill delays in their preschool years had language skills that were not significantly different from their peers by second grade. Using standardized measures of language assessment, these studies have demonstrated that the basic language skills of children with a history of otitis media, when other factors are considered, are not negatively affected in the long-term.

There is a substantial body of work that does find a relationship between early and prolonged exposure to otitis media and language related skills when measures other than standardized measures are employed. Teele et al. (1990) found that the only language skill that was significantly different in children with a history of otitis media was their use of morphological markers, while the results of the standardized vocabulary assessments did not reveal differences in skill level. The work of Vernon-Feagans and colleagues has often focused on information beyond that of standardized language tests in order to determine if there are differences in the social use of language. Vernon-Feagans, Kipp, and Blood (1994) investigated the interaction between well children and those with chronic otitis media and the impact of low- versus high-quality daycare centers on language development. They found no differences in language skills at 24 months, although differences did emerge in terms of attention skills when a history of otitis media was coupled with low quality daycare. In a further investigation of the interaction between otitis media and the environmental variable of quality of daycare on language development in children, Vernon-Feagans, Emanuel, and Blood (1997) examined the performance of 24 month old children on a comprehensive assessment of receptive and expressive language. Differences were again noted in relation to quality of daycare and chronic otitis media, although in this study children with chronic otitis media who were in low-quality daycare had significantly lower scores on the expressive portion of the assessment. Continuing to examine this interaction between environmental factors, otitis media, and language related skills, Vernon-Feagans, Hurley, and Yont (2002) examined mother/child book reading activities in 4 year olds. The results of this study indicated that children with chronic otitis media in low-quality daycare had greater difficulty producing correct answers in the book reading activity, their responses contained shorter MLU, fewer different words, and fewer total words when compared to the other groups of children. Finally, Creps and Vernon-Feagans (2000) examined the effects of otitis media and daycare on child developmental factors such as language and social interaction. The results of this study indicated that the best predictor of language skills at 7 years of age was a combination of a history of otitis media, the age of entry into daycare, and the quality of daycare. The cumulative work of Vernon-Feagans and colleagues demonstrates that while long term effects of otitis media may not always be evidenced through standardized assessments, methods that look at how the child is functioning in the environment do show differences in language skill use and attentional skills.

At first glance, the results of these studies may appear to be in conflict with each other, as some studies have demonstrated no differences in language related skills in association with a history of otitis media, while others have. These studies do provide evidence that the hearing status and presence of mild hearing loss and/or otitis media does influence language outcomes at least on occasion and should be considered when selecting young children as participants for research related to language development.

Risk of Otitis Media in Children with Clefts

Children with clefts are at higher risk for middle ear infections than children without clefts (Handzic-Cuk, Cuk, Risavi, Katusic, & Stajner-Katusic, 1996; Sheahan, Miller, Sheahan, Earley, & Blayney, 2003; Valtonen, Dietz, & Qvarnberg, 2005). Eustachian tube dysfunction is the primary reason for increased middle ear infections, as the muscle that is most responsible for opening the Eustachian tube, the tensor veli palatini, has an abnormal course and insertion in children with cleft palates (Broen et al., 1996). Studies investigating long-term effects of middle ear disease in children with clefts frequently differ in terms of recommendations for the insertion of ventilation tubes, as some argue against repeated placement of ventilation tubes as they lead to long term hearing loss (Sheahan, Blayney, Sheahan, & Earley , 2002; Sheahan et al., 2003), while others question if the persistent hearing loss is secondary to the effects of long term otitis media with effusion (Goudy, Lott, Canady, & Smith, 2006). The mangagement of otitis media in children with clefts remains variable as a result of the conflicting findings associated with these studies.

Given the risks associated with otitis media, hearing loss and delayed language development, it is somewhat surprising to note that hearing status is not uniformly noted in studies investigating language development in children with clefts. The trend seems to be that while hearing status is reported in terms of passing a screening at the time of a speech and language evaluation, little consideration is given to ongoing episodes of middle ear infections, presence of ventilation tubes, number of ventilation tubes or hearing acuity over time. In the earlier discussion of language development in infants and toddlers with history of repaired cleft palate, two studies did not report findings associated with hearing status (Starr et al., 1977; Neiman & Savage, 1997). Speltz et al. (2000) indicated records were obtained to determine the number of times children were treated by their physician for middle ear infections, but included this information as part of a larger maternal factor in relation to their language findings and did not conduct an analysis based on hearing acuity. Several other studies included hearing status in the analysis, but did not find significant differences that impacted language skills (Chapman et al., 2003; Fox et al., 1978; Jocelyn et al., 1996). However, Broen et al. (1998) noted that it was hearing status that accounted for much of the variance in language scores in infants and toddlers with and without cleft palate. In comparison to the infant/toddler studies, hearing status was often discussed in studies investigating language development in preschool/school age children with clefts. Most studies, six studies out of seven, at least made note of a hearing screening that was used to exclude children who did not demonstrate adequate hearing thresholds on the day(s) of the study; however, the cut-off level was inconsistent across studies, ranging from 20 dB to as high as 35 dB loss. Given the ongoing discussion relating to the impact of otitis media and hearing loss on language development, it is important for researchers to include threshold testing information on their subjects in addition to data on number of otitis media episodes and consider these factors in relation to language outcomes.

Summary of Converging Literature

As demonstrated through this review of the literature, PPA skills, speech impairments, and language skills converge when an individual is learning to read. Children with clefts are known to face significant difficulties with speech production skills in their early years as a result of their oral structural deficits. Their receptive and expressive language skills are at risk for being mildly delayed. However, their phonological awareness skills have yet to be explored, and their reading skills have not been explored extensively. Given the convergence of known risk factors, it is important to investigate these skills more comprehensively.

Literacy Development of Children with Clefts

Children with clefts are at risk for developing reading difficulties, perhaps as a result of the risk factors already explicated. Richman, Eliason, and Lindgren (1988) reported that in a sample of children with cleft lip and palate and isolated cleft palate, 35 percent had a moderate reading disability and 17 percent had a severe disability. They also analyzed their data on the basis of cleft type, indicating that 48% of their sample of children between the ages of 6 and 7 years with cleft lip and palate demonstrated some form of reading disability; this is similar to 53% of the children in the group with isolated cleft palate that had reading disability at that age. By age 10 to 13 years, however, the percentage of children with cleft lip and palate with reading disability had decreased to 8.6%, which is reflective of the rate of reading disability in the general population. Conversely, the impact of isolated cleft palate on reading skills appears to have a longer lasting effect, as the percentage of reading disability only decreased to 33% at age 10-13 years, which is significantly higher than the general population. Richman et al. (1988) indicate that the high rate of reading disability at the 6 to 7 years of age marker for both children with cleft lip and palate as well as those with isolated cleft palate is possibly an interaction between the speech production difficulties that children with clefts continue to face at this age and the reading skills that are being addressed such as sound-symbol relationships and syllable blends.

In a more recent study, Richman and Ryan (2003) sought to identify the type of reading disability seen in the cleft lip and palate population. The subjects in this study ranged in age from 7 to 11 years old and all were diagnosed as having nonsyndromic cleft lip and palate. After completing a battery of tests that included sound blending tests, word fluency tests, and rapid naming activities, the authors concluded that the children with cleft lip and palate had deficits in rapid naming and relative strengths in PPA. According to Richman and Ryan, this strength in PPA skills suggests that effective reading instruction for this population would employ phonics based approaches embedded in meaningful texts.

While practitioners working with children with clefts often share anecdotal evidence that children with clefts are at risk for reading disabilities, studies that have examined reading skills in this population are limited. The study by Richman and Ryan (2003) was the only study identified that attempted to describe the reading difficulties facing children with clefts. Richman and Ryan indicated that phonemic awareness skills were a relative strength; however, the impact of the broader set of phonological skills on their reading abilities was not discussed. Also, language was not assessed in terms of syntax and morphology; the contribution of those skills to the overall reading abilities of children with clefts is also unknown.

In conclusion, individuals with clefts are at a higher risk for reading disability than the general population, at least in the early elementary years. The relationship between disordered phonological systems, language impairment, and PPA skills has been established in the literature, as has the risk of delay or disorder in one or more of these areas in children with clefts. Based on the fact that children with clefts often exhibit significant speech disorders in their early years secondary to their structural defects and their risk for mild language delays, the status of PPA skills and language skills of young children with clefts warrants further investigation.

CHAPTER 3

Methods

The purpose of the investigation reported herein was to explore the phonological awareness skills of a group of children with and without clefts, ranging in age from 4 years 0 months to 6 years 11 months old.

Research Questions

One primary and three secondary research questions drove the investigation. Based on the hypothesis that the phonological awareness skills of children with clefts would be lower than those of the general population, the primary research question was:

1. Are the phonological awareness skills of 4 to 6 year old children with clefts different from those of the general population?

As it is currently understood that children with clefts in this age range often have speech production errors that are severe in nature, it follows that they may have phonological awareness skills that differ from the general population. The three secondary questions were intended to explore factors that may contribute to a difference in phonological awareness skill performance of children with clefts. The three secondary research questions were:

1. Is there a relationship between speech production and phonological awareness skills in children with clefts?

It was hypothesized that children with decreased phonological awareness skills would have a higher number of speech production errors.

2. Is there a relationship between language skills and phonological awareness skills

in children with clefts?

It was hypothesized that the contribution of oral language skills in the development of phonological awareness skills would lead children with decreased language skills to have decreased phonological awareness skills (Cooper, Roth, Speece, & Schatschneider, 2002).

3. Is there a relationship between hearing status and phonological awareness skills in children with clefts?

It was hypothesized that children with histories of increased otitis media and fluctuating hearing loss would have decreased phonological awareness skills.

Participants

Forty-six children, 23 with clefts and 23 without clefts, participated in this study. Both boys and girls were eligible for this study. All ethnic groups were invited to participate, although children in the study were required to use English as their dominant language given that the language assessments were all in English and the normative data were for English speaking children.

Characteristics of Children with Clefts

To qualify for participation in this study, the children with clefts met the following criteria:

1. Children must have a history of unilateral complete cleft of the primary and secondary palate, cleft palate only, or bilateral cleft lip and palate.
2. Children must not have any known syndromes, cognitive impairments, or neurological deficits.
3. Children with clefts must have had palatal repair performed according to the surgical guidelines recommended by the American Cleft Palate-Craniofacial Association or following

the standard protocol for lip and palate repair at Texas Children's Hospital, where the children with clefts were recruited for participation in this study. The American Cleft Palate-Craniofacial Association (2002) indicates that children have lip closure surgery at approximately 2-3 months, with surgery to close the palate ranging between 6 to 18 months of age. At Texas Children's Hospital, the standard protocol for lip and palate repair often includes an additional lip closure procedure called a lip adhesion that brings the edges of the cleft together without attempting to close the defect in its entirety; when indicated, this is the first surgery to occur and usually is completed between 5 to 9 months of age. Following the lip adhesion procedure, children at Texas Children's Hospital generally had a definitive lip repair between 9 and 12 months of age. Finally, the cleft palate is routinely repaired between 12 and 15 months of age. If a child had a cleft palate only, the palate was typically repaired between four and nine months of age. Table 1 provides specific information for each participant with a cleft regarding the age at which necessary repairs were completed.

4. Children must have chronological age between 4;0 and 6;11 years of age at the time of participation for the study.

5. Children must use English as their dominant language.

6. Children must have sufficient hearing on the day of testing (with hearing thresholds of at least 35dB at 500Hz, 1K, 2K, and 4KHz in either ear) to rule out hearing loss as the primary cause of poor performance on the phonological awareness and language assessments administered; this screening level is consistent with other studies being conducted in preschool settings where ambient noise is likely to be high (Plante & Vance, 1995).

Table 1

Cleft Type and Age at Lip and Palate Repair Procedures

Participant	Cleft type	Lip adhesion	Definitive lip repair	Palate repair
11ev10ur	CPO[1]	n/a	n/a	7 months
1le1rr	CPO	n/a	n/a	5 months
22in1rr	CPO	n/a	n/a	5 months
11ay8ol	CPO	n/a	n/a	6 months
23il6ob	CPO	n/a	n/a	6 months
11ea6ri	CPO	n/a	n/a	4 months
13ad5sq	CPO	n/a	n/a	9 months
11en7ra	UCL&P[2]	n/a	2 ½ months	11 months
10am19ut	UCL&P	n/a	3 months	6 months
3ol3ir	UCL&P	n/a	2 ½ months	10 months
12uc11no	UCL&P	n/a	5 months	10 months
13ad8on	UCL&P	n/a	3 months	13 months
1na13or	UCL&P	n/a	8 months	15 months
26ac19mi	UCL&P	n/a	3 months	8 months
3hr3as	UCL&P	6 months	9 months	13 months
5ri19pe	UCL&P	5 months	8 months	11 month
10oh2al	UCL&P	5 months	9 months	12 months
10ac3an	UCL&P	6 months	9 months	12 months

[1] CPO indicates Cleft Palate Only
[2] UCL&P indicates Unilateral Cleft Lip and Palate

23ya18ad	BCL&P[3]	9 months	12 months	16 months
20ro12in	BCL&P	n/a	2 ½ months	12 months
19av10oh	BCL&P			
20ri23al	BCL&P	7 months	11 months	18 months
19av20hi	BCL&P	6 months	9 months	12 months

In order to complete the planned statistical analysis for the primary question, a power analysis using G-Power (Erdfelder, Faul & Buchner, 1996) suggested that a total sample size of 46 (with 23 in each group) would have 85% power to detect a difference in means of 10.000, assuming a standard deviation of differences of 15, using a paired t-test with a 0.050 two-sided significance level.

Characteristics of Children in Matched Comparison Group

Participants in the comparison group were matched with those in the experimental group according to age, gender, race, mother's education level, and school experience. Detailed demographic information is provided in Table 2, with matched pairs listed next to one another.

[3] BCL&P indicates Bilateral Cleft Lip and Palate

Table 2

Characteristics of Matched Pairs

Pair #	Participant	Presence of cleft	Age (in months)	Gender	Ethnicity	Mother's education level	Participant's school experience
1 a	11en7ra	Yes	56	Female	African American	Grad Sch[4]	Pre-K
b	12ei7ra1	No	52	Female	African American	Grad Sch	Pre-K
2 a	1na13or	Yes	50	Female	Hispanic	High Sch[5]	No school
b	7ab7ib1	No	53	Female	White	4-Y Coll[6]	No school
3 a	10ac3an	Yes	59	Female	Hispanic	High Sch	No school
b	1is2ar1	No	59	Female	Hispanic	4-Y Coll	Pre-K
4 a	13ad5sq	Yes	59	Female	Hispanic	High Sch	Pre-K
b	6ra5sq1	No	59	Female	Hispanic	High Sch	Pre-K
5 a	19av10oh	Yes	61	Female	Hispanic	High Sch	Pre-K
b	10ai3ar1	No	62	Female	White	Grad Sch	Pre-K
6 a	13ad8on	Yes	55	Female	White	High Sch	No school
b	3as4on1	No	56	Female	White	High Sch	Pre-K
7 a	11ea6ri	Yes	63	Female	White	High Sch	No school
b	3he12ak1	No	71	Female	White	High Sch	Kindergarten

[4] Grad Sch= Advanced Degree/Graduated from Graduate School
[5] High Sch. = High School Graduate
[6] 4-Y Coll. = Four Year College Graduate

8 a	19av20hi	Yes	75	Female	White	High Sch	First Grade
b	19he11in1	No	74	Female	White	High Sch	First Grade
9 a	11ay8ol	Yes	75	Female	White	High Sch	Kindergarten
b	2ai6lo1	No	76	Female	White	No H.S.[7]	First Grade
10 a	3hr3as	Yes	50	Male	Hispanic	Grad Sch	No school
b	5th18od1	No	54	Male	Hispanic	High Sch	Pre-K
11 a	23il6ob	Yes	48	Male	White	4-Y Coll	Pre-K
b	19et5if1	No	48	Male	White	4-Y Coll	No School
12 a	1le1rr	Yes	51	Male	White	High Sch	Pre-K
b	1le10oh1	No	53	Male	White	High Sch	Pre-K
13 a	22in1rr	Yes	51	Male	White	High Sch	Pre-K
b	18ei16al1	No	55	Male	White	High Sch	Pre-K
14 a	23ya18ad	Yes	55	Male	White	High Sch	Pre-K
b	18il19ug1	No	58	Male	White	High Sch	Pre-K
15 a	11ev10ur	Yes	62	Male	White	High Sch	Pre-K
b	4yl2is1	No	60	Male	White	High Sch	Pre-K
16 a	20ri23al	Yes	66	Male	White	No H.S.	No school
b	19te13or1	No	61	Male	White	High Sch	Pre-K
17 a	3ol3ir	Yes	72	Male	White	High Sch	Kindergarten
b	2ry23il1	No	62	Male	White	High Sch	Pre-K
18 a	10oh2al	Yes	77	Male	White	4-Y Coll	Kindergarten
b	20om1da1	No	73	Male	White	High Sch	First Grade

[7] No H.S. = Did not Graduate High School

19 a	26ac19mi	Yes	79	Male	White	High Sch	Kindergarten
b	2ra4o1	No	76	Male	Hispanic	High Sch	Kindergarten
20 a	20ro12in	Yes	80	Male	White	High Sch	First Grade
b	10ac12ar1	No	77	Male	White	High Sch	First Grade
21 a	5ri19pe	Yes	80	Male	White	4-Y Coll	Kindergarten
b	3ha16al1	No	79	Male	White	High Sch	First Grade
22 a	12uc11no	Yes	82	Male	White	4-Y Coll	First Grade
b	10ar23il1	No	83	Male	White	High Sch	First Grade
23 a	10am19ut	Yes	83	Male	White	4-Y Coll	Kindergarten
b	5ri5if1	No	83	Male	White	4-Y Coll	First Grade

Recruitment and Matching Procedures

All participants in the cleft group were recruited through the Cleft Palate Clinic at Texas Children's Hospital in Houston, TX. Texas Children's Hospital is a large urban pediatric hospital with a high volume cleft palate clinic. Texas Children's Hospital is a training site for doctors in the Plastic Surgery Residency Program at Baylor College of Medicine. One primary plastic surgeon completes the cleft lip and palate repairs for children who are seen by this team.

Participants in the comparison group were recruited through a number of resources. A presentation was made to the Department of Speech, Language, and Learning in order to inform colleagues about the study in an effort to identify friends and family members of colleagues who had or knew children who fit the age requirements. Flyers (see Appendix B) were distributed to colleagues, affiliated doctors' offices, and friends in an effort to recruit typically developing children. Most participants in the comparison group were family members (children, nieces, and nephews) of individuals working at Texas Children's Hospital. Several daycare center directors also allowed the primary investigator to come to their daycare centers to recruit and test children for the study.

If interested, parents of participants contacted the primary investigator by phone or email. Once contact had been made, the study was discussed with the volunteers. If the volunteers were still interested in participating, demographic information was gathered over the phone (see Appendix C for Demographic Form). The demographic data obtained via phone interview included identifying information for the child, language spoken at home, type of cleft (if applicable), gender and ethnicity of the child, and mother's level of education. In addition, information regarding hearing history and school experience was gathered using this interview. For example, when reporting hearing history, parents were asked questions to recall the number of times their child had been treated for middle ear infections over the last two years. Specifically, they were asked to indicate which choice best described their child's experience: 0 ear infections, 1 to 3 ear infections, 4 to 6 ear infections, or 7 or more ear infections within the last two years. A follow-up question was then asked in order to determine if this number had increased, decreased, or remained consistent over the years. Parents were also asked if their child had PE tubes, and if so, how many sets had been inserted and the age which they were first placed. The child's involvement in special services including speech-language therapy, physical therapy, occupational therapy, or any other special education service was recorded along with information regarding the child's preschool and school experience.

If the participant had a cleft, an appointment was then made for the child and a parent to come to Texas Children's Hospital for the assessment. If the potential participant was going to be in the comparison group, the demographic information was then compared with the participants with clefts and a determination regarding matching status was made. If a comparison child matched well with a child from the group of children with clefts, an appointment was made for the child and a parent to come to the assessment. If the child did not match with a child in the group of children with clefts, their contact information was retained until the end of the study in case a match was identified at a later time. If a match was not immediately identified from the list of participants with clefts, a parent volunteering her child for the comparison group was told that she would only be contacted in the future if a match was identified; if no match was identified, her contact information would be destroyed. All participants received a written report detailing the results of testing as well as any recommendations for intervention, as needed.

Procedures

Description of the Assessment Session

All children with clefts were tested at Texas Children's Hospital in private treatment rooms. Eleven of the 23 children in the comparison groups were tested at Texas Children's Hospital in similar private treatment rooms. The remaining twelve participants in the comparison group were tested at their daycare centers. Efforts were made to test the children in quiet rooms that were free of visual and auditory distractions in their daycare centers.

When the child and his/her parent arrived at the assessment location, the informed consent documents were reviewed. In cases where testing was completed at daycare centers, written consent was obtained from the child's parent prior to initiating testing. Given that the sample consisted of children ranging in age from 4;0 to 6;11 years old, the child's parent was asked to sign the informed consent form. Child assent was not required. After completing the required consent documents, the parent left the room.

Hearing Screening Procedures

Prior to beginning the test protocol, each child's hearing was screened using a Beltone Audiometer Model 10D that was calibrated annually. At the onset of the study the last calibration had been in February 2005, and the audiometer was calibrated for the second time in September 2006. A hearing screening was conducted at 35 dB at 500, 1-, 2-, and 4-K Hz. A level of 35 dB was set based on anticipated level of ambient noise in the room and is consistent with a screening level used by other researchers investigating language development in preschool-aged children (Plante & Vance, 1995; Gray, Plante, Vance, & Henrichsen, 1999). Those who did not pass a hearing screening were referred to an audiologist as well as their otolaryngologist for further testing and treatment. Children continued with the test protocol as long as the hearing screening was passed in at least one ear, as the child was determined to have sufficient hearing to continue with speech and language testing. Several children appeared to have difficulty training to task for the hearing screening. In these cases, the tones were presented several times to ensure that a failure to indicate hearing a particular tone was reflective of a hearing loss, rather than difficulty with the screening task. Results of the hearing screening were marked on the Lesson Plan (see Appendix D).

Order of Administration

Following the hearing screening, the children in the study completed the three or four tests depending on their age. The tests were completed in a relaxed environment, with both the researcher and the child participant seated at a child-sized table. The various tests were administered by the researcher who is a licensed speech language pathologist familiar with the speech patterns that are typical in children with cleft lip and palate. The tests included a test of phonological awareness, The Pre-Reading Inventory of Phonological Awareness (PIPA, Dodd, Crosbie, McIntosh, Teitzel, and Ozanne, 2003); a test of language competence, The Clinical Evaluation of Language Fundamentals-Preschool-2nd Edition (CELF-P:2, Wiig, Secord, & Semel, 2004); a test of speech production, The Goldman-Fristoe Test of Articulation-2 (GFTA-2, Goldman & Fristoe, 2000); and a test of oral reading fluency for the participants enrolled in first grade, the Dynamic Indicators of Basic Early Literacy Skills (DIBELS, Good & Kaminski, 2002). The tests were administered in the following order: (a) CELF-P:2, (b) PIPA, (c) GFTA-2, and (d) DIBELS. A snack consisting of juice and cookies or crackers was offered after the CELF-P:2. If the child was not interested in a snack break, they were given the option of coloring or playing with figurines.

Total Length of Session

In every case, testing was completed during one visit, although protocol allowed for testing to be completed over two days if needed in order to for the child to provide adequate attention to task. Testing was completed in a total of 1 to 3 hours.

Description of the Measures

Phonological Awareness Measures

The Pre-Reading Inventory of Phonological Awareness (PIPA; Dodd, Crosbie, McIntosh, Teitzel, and Ozanne, 2003) was used to assess phonological awareness skills. The PIPA has been shown to be reliable and valid in testing phonological skills in children as young as age 4 and has an upper age limit of 6;11. According to data provided in the PIPA manual, test-retest stability calculated with Pearson's product-moment correlation for the individual subtests of the PIPA are as follows: Rhyme Awareness--.80; Syllable Segmentation--.67; Alliteration Awareness--.83, Sound Isolation--.94; Sound Segmentation--.83, and Letter-Sound Knowledge--.97 (Dodd et al. 2003). Reliability using the Split-Half Method based on subtests and six-month age categories ranged from .68 to .98 (Dodd et al., 2003). These values suggest that reliability of this measure is good. The authors of the PIPA note that the subtests demonstrate content validity, in that the tasks in PIPA are predictive of risk of reading failure and have been shown to accurately identify weaknesses (Dodd et al., 2003).

The PIPA (Dodd et al., 2003) covers six different skills associated with phonological awareness. The test was administered according to the guidelines set forth in the examiner's manual. Each subtest was administered. All subtests began with a demonstration and trial items. One repetition was allowed for all but one of the subtests if the child requested repetition or did not respond; the Letter-Sound Knowledge subtest did not allow repetition. The child was given one point for each correct answer; raw scores were then converted to percentile ranges for each subtest. The specific subtests are described below:

1. Rhyme awareness assessed the child's ability to determine which word does not rhyme with the others from a field of four. The speech-language pathologist said each word while pointing to a corresponding picture from the stimulus booklet that was available to help the child remember the words. The child indicated the word that did not rhyme with the others.

2. Syllable segmentation assessed the child's ability to break down words into syllables. The speech-language pathologist said a word that ranges from two to five syllables and the child segmented the word.

3. Sound segmentation was similar to syllable segmentation, but rather than separating words into syllables, the child was required to divide the word into individual sounds or phonemes. The speech-language pathologist said the word, and the child segmented the word into phonemes.

4. Alliteration awareness examined the child's ability to determine which word in a group of four words began with a different sound. For this subtest, the speech-language pathologist pointed to pictures as they were named, and the child was required to identify which word did not begin with the same sound. The pictures were provided to help the child remember the words.

5. Sound isolation assessed the child's ability to identify the onset of a word and label it. The speech-language pathologist said a word and the child was required to say the first sound in that word.

6. Letter-Sound knowledge required the child to look at a printed letter and produce the sound it makes.

Language Measures

The Clinical Evaluation of Language Fundamentals-Preschool-2nd Edition (CELF-P:2) (Wiig, Secord, & Semel, 2004) was used to assess language skills. The CELF-P:2 was selected as the language measure to be used in this study because of its strong psychometric properties and the variety of language skills assessed using this measure. According to the examiner's manual, the average across ages test-retest reliability for the various subtests of the CELF-P:2 ranged from .78 to .90. Reliability based on the Split-Half Method for the different age groups and subtests ranged from .61 to .97. The average reliability was greater than .90 for composite scores (Wiig, Secord, & Semel, 2004). According to Wiig, Secord &

Semel (2004) content validity was determined by review by a panel of experts and by testing the correlation between the CELF-P:2 and the CELF-P (Wiig, Secord, & Semel, 1992), the CELF-4 (Semel, Wiig, & Secord, 2003), and the PLS-4 (Zimmerman, Steiner, & Pond, 2002). These comparisons revealed moderate correlations between the CELF-P:2 and the CELF-P and the PLS-4, and a moderate to high correlation between the CELF-P:2 and the CELF-4.

The CELF-P:2 (Wiig, Secord, & Semel, 2004) was suitable for the age range included in this study, as it was standardized for ages 3;0 to 6;11. The CELF-P:2 was administered in its entirety according to the guidelines set forth in the examiner's manual. Seven subtests were administered. The subtests were as follows:

1. Sentence Structure examined the child's ability to understand sentences of different lengths and complexity.
2. Word Structure evaluated the child's ability to use various morphological markers and pronouns appropriately.
3. Expressive Vocabulary examined the child's ability to label people, objects, and actions.
4. Concepts and Following Directions examined the child's ability to follow directions of varying complexity and lengths.
5. Recalling Sentences examined the child's ability to listen to sentences and repeat them without altering meaning or syntax.
6. Basic Concepts tested the child's ability to understand one-step directions containing various modifiers.
7. Word Classes evaluated the child's ability to group words according to semantic class and express that class orally.

Speech Production Measure

The Goldman-Fristoe Test of Articulation-2 (GFTA-2, Goldman & Fristoe, 2000) was used to assess speech production. The GFTA-2 is commonly used to assess speech production. The Sounds in Words subtest consists of stimulus pictures that are used to elicit target words, which were then transcribed using narrow transcription, including diacritics to mark distortions due to dentition and lateralization of phonemes that are commonly observed in children with clefts. The Sounds in Words subtest was scored according to the guidelines in the examiner's manual; standard scores and percentile ranks were obtained.

Literacy Measure

The Oral Reading Fluency (ORF) subtest of the Dynamic Indicators of Basic Early Literacy Skills (DIBELS, Good & Kaminski, 2002) was used to assess accuracy and fluency of reading in connected text. This measure was only given to children in either the experimental or comparison group who were enrolled in first grade. This standardized measure consists of three reading passages that the child reads aloud. The total number of words read aloud within one minute was calculated according to the administration guidelines; words were counted as incorrect if they were omitted, spoken in error, or followed hesitations lasting more than three seconds, while words that were self-corrected within three seconds were scored as correct. The median score from the three passages was used to determine the oral fluency rate. The child's performance was categorized as at risk (less than eight words read correctly), some risk (between eight and 20 words read correctly), or low risk (greater than 20 words read correctly) (Good, Kaminski, Simmons, & Kame'enui, 2001).

Data Management

All notes, tests, and other records were stored in a locked cabinet in a private office at Texas Children's Hospital in order to ensure privacy. Participants were assigned identification codes rather than having identifying information on these records. The methods as described herein were initially approved by the Institutional Review Board of the Medical School at the University of North Carolina and then later approved by the Institutional Review Board at Baylor College of Medicine.

Inter-Rater Reliability

Secondary scoring for reliability purposes was completed by a second licensed speech language pathologist who also was familiar with the speech patterns that are typical in children with cleft lip and palate. Sessions were audio taped for reference when discrepancies were noted between scorers. Inter-rater reliability measures were conducted for 30% of all of the assessments completed. Sessions that were co-scored were randomly selected throughout the course of the study.

When a child's assessment was co-scored, the primary investigator and the child were seated at a child-sized table. The second speech language pathologist was present during the assessment, but sat away from the table in order to minimize any further distractions. Each speech-language pathologist scored the test independently. The total scores were compared for accuracy. When there was a discrepancy between the two scores, the audiotape was played for clarification while both speech-language pathologists independently transcribed the response. The transcriptions were again compared. If discrepancies remained, errors were discussed and a final decision was made with mutual agreement.

Point-by-point interrater reliability was calculated using the total number of agreements divided by the number of agreements plus disagreements between the two scorers. Interrater reliability for individual subtests of the PIPA ranged from 99.3% to 100%, with the overall reliability at 99.6%. Interrater reliability for individual subtests of the CELF-P:2 ranged from 88.6% to 100%, with overall interrater reliability of the CELF-P:2 at 98.7%. Interrater reliability for the GFTA-2 was 96.0%.

Plan for Analysis

Several statistical analyses were conducted to address the questions driving the research. Across the multiple analyses described below, Type I error was a concern. As this was the first study of its kind, the information that results from the analyses contributes to what is known about children with clefts, but should be interpreted with caution, as corrections to control for Type I error were not employed. If corrections such as Bonferroni had been employed, no significant findings would have resulted and little guidance would be available for future investigations. While corrections were not made, efforts were taken to restrict Type I errors. For example, analyses were planned whenever feasible given the existing literature and alpha was set at .01. When findings that were significant at the .05 level are reported, these findings should be considered with more caution than those at the .01 level, as they have a greater likelihood of showing an effect that is actually due to chance. The analysis procedures that were employed are described below with reference to each question.

1. Are the phonological awareness skills of 4 to 6 year old children with clefts different from those of the general population?

A two-tailed paired samples t-test was used to compare the performance of children with clefts to their peers on each of the six subtests of the PIPA. Secondly, based on each subtest from the PIPA, one sample t-tests comparing the sample population mean to the normed population mean was analyzed using SPSS 14.0 for Windows.

a. Is there a relationship between speech production and phonological awareness skills in children with clefts?

Pearson product moment correlation was used to conduct this analysis comparing performance on each of the subtests of the PIPA with performance on the GFTA-2.

b. Is there a relationship between language skills and phonological awareness skills in children with clefts?

Pearson product moment correlation was used to conduct this analysis comparing performance on each of the subtests of the PIPA with overall performance on the CELF-P:2.

c. Is there a relationship between hearing status and phonological awareness skills in children with clefts?

Multiple regression was planned to analyze the relationship between performance on the total PIPA with results of the hearing screening, amount of time spent during the first years of life with otitis media, and the frequency of otitis media during the two years prior to participation in the study. With the exception of the scores on the PIPA, scores in the multiple regression reflected the parent responses to multiple choice questions on the parent interview.

Summary of Methods

This study was designed to evaluate the phonological awareness skills of children with clefts ranging in age from 4 to 6 years old and compare them to a group of matched peers without clefts as well as normative data available. Using commercially available assessments of phonological awareness, speech production, and language skills, the skills of 23 children with clefts were assessed and compared to their peers and to normative data. Further analysis was planned to investigate relationships between phonological awareness, speech production, oral language skills, and hearing status. When appropriate, performance on oral reading fluency was also to be compared.

CHAPTER 4

Results

Multiple analyses were conducted in order to test the four hypotheses that drove the research questions in the current investigation. These analyses are described below with reference to each of the research questions.

Sample Characteristics

Prior to beginning the planned analyses, demographic data were first explored using frequency tables (Table 3) to investigate the similarities between the two groups. The children in the two groups were similar on four out of the five planned matching variables. Age, gender, ethnicity, and mother's education were closely matched between the two groups. Based upon parent report, school experience was not closely matched, with a higher number of children in the comparison group participating in structured education; however, when collecting the data for the school experience variable, it became clear that parents of children who were not yet in kindergarten or first grade did not consistently report a child's involvement in formal school. For example, when obtaining information from parents who had children enrolled in the same full-day daycare center, some children who did not attend an outside pre-kindergarten or preschool were reported as being in pre-kindergarten while others were said to not be in school. For that reason, it is difficult to determine if there are true group differences with respect to school experiences or if the differences merely reflect differences in parent perception of what it means for a young child to be enrolled in school versus childcare.

Table 3

Demographic Characteristics of Cleft and Comparison Groups

	Number (%) of Children		
Characteristic	Cleft	Comparison	Total
Total Children	23	23	46
Gender			
Male	14 (60.9)	14 (60.9)	28
Female	9 (39.1)	9 (39.1)	18
Ethnicity			
African American	1 (4.3)	1 (4.3)	2
Hispanic or Latino	5 (21.7)	4 (17.4)	9
White, not Hispanic	17 (73.9)	18 (78.3)	35
Mother's Education			
Did not complete high school	1 (4.3)	1 (4.3)	2
High school graduate	15 (65.2)	16 (69.6)	31
Four year college graduate	5 (21.7)	4 (17.4)	9
Graduate school	2 (8.7)	2 (8.7)	4
Year in school			
Not in school	6 (26.1)	2 (8.7)	8
Pre-Kindergarten	8 (34.8)	12 (52.2)	20
Kindergarten	6 (26.1)	2 (8.7)	8
First grade	3 (13.0)	7 (30.4)	10
Age			

Mean (months)	64.7	64.5
Range (months)	48 – 83	48 – 83

The data were then screened at the univariate level to insure that they met the assumptions of the planned analysis. Specifically, the normality of the data was tested with respect to its skewness and kurtosis. As shown in Table 4, the participant percentile scores on the PIPA subtests were normally distributed based on skewness and kurtosis statistics for both the cleft and comparison groups. Similarly, composite measures of the CELF-P:2 were normally distributed for both groups. While the statistics for the children with clefts were normally distributed on the GFTA-2 , the comparison group was negatively skewed. Given that entrance criteria for children in the comparison group were not to be enrolled in speech therapy, the finding that this group had a high number of above average scores is to be expected. Based on these results, data analysis continued as planned.

Phonological Awareness

Group Comparisons

For each subtest of the PIPA, raw scores were converted to percentile ranges in five point increments from 0 to 99, which were then given a value from 1-20 in order to conduct further analyses without using a range. For example, a raw score that was converted to a percentile range of 0-4 using the PIPA manual was given a value of 1. Similarly, a raw score that was converted to a percentile range of 5-9 using the PIPA manual was then given a value of 2, and so forth. Group mean and standard deviations were computed using scores from the percentile ranges. The resulting paired samples statistics for each subtest are displayed in Table 5. A paired-samples t-test was then conducted for each subtest on the PIPA to evaluate whether children with clefts had phonological awareness skills that were different from children without clefts. As shown in Table 6, results indicated that mean scores for each subtest did not differ significantly between the two groups.

Table 4

Skewness and Kurtosis Statistics for Cleft and Comparison Group Performance on PIPA, CELF-P:2, and GFTA-2

	Skewness z-score		Kurtosis z-score	
	Cleft	Comparison	Cleft	Comparison
PIPA subtest				
Rhyme Awareness	0.21	1.53	1.26	0.90
Syllable Segmentation	0.26	0.60	1.18	0.97
Alliteration Awareness	0.52	1.05	1.28	0.97
Sound Isolation	0.26	-0.56	1.27	1.32
Sound Segmentation	0.58	0.85	1.04	1.26
Letter-Sound Knowledge	-1.15	-0.12	1.19	1.30
CELF-P:2 Composite Score				
Core Language	0.39	1.13	0.45	0.95
Receptive Language	-1.00	0.34	0.36	0.88
Expressive Language	-0.18	0.88	0.44	0.46
Language Content	-0.54	0.83	0.37	0.75
Language Structure	-0.69	0.41	0.55	0.74
GFTA-2	-0.12	-3.97	0.90	2.33

Table 5

Paired Samples Statistics for PIPA Subtests

Subtest	Mean Converted Scores	Range*	Standard Deviation
Rhyme Awareness			
Cleft	9.22	1-18	5.85
Comparison	7.13	1-19	5.93
Syllable Segmentation			
Cleft	10	1-18	5.53
Comparison	9.35	1-18	4.96
Alliteration Awareness			
Cleft	9.39	1-19	6.28
Comparison	9.04	1-19	5.52
Sound Isolation			
Cleft	10.43	3-19	5.11
Comparison	11.52	3-19	6.11
Sound Segmentation			
Cleft	11.22	5-20	4.56
Comparison	9.87	2-20	5.89
Letter-Sound Knowledge			
Cleft	12.43	3-19	5.51
Comparison	11.35	3-20	6.12

* each subtest has a maximum score of 20

Table 6

Paired Samples Tests for Converted Scores on PIPA Subtests—Paired Differences

Subtest	Mean	Standard Deviation	95% Confidence Interval of the Difference		t	Significance (2-tailed)
			Lower	Upper		
Rhyme Awareness	2.09	7.97	-1.36	5.53	1.26	0.22
Syllable Segmentation	0.65	6.73	-2.26	3.56	0.47	0.65
Alliteration Awareness	0.35	7.43	-2.86	3.56	0.23	0.82
Sound Isolation	-1.09	7.41	-4.29	2.12	-0.70	0.49
Sound Segmentation	1.35	6.51	-1.47	4.16	0.99	0.33
Letter-Sound Knowledge	1.09	6.07	-1.54	3.71	0.86	0.40

Comparisons with Normative Data

A one-sample t-test was conducted on each of the PIPA subtests in order to evaluate whether the children with clefts earned a mean score that was significantly different from the normative data provided in the PIPA manual. A t-test, rather than a z-test, was used given the sample size of 23, as the t-test is to be used when $n<30$ (Blaisdell, 1993). Since the PIPA manual only provides percentile scores in 5-point ranges, the scores for the participants had to be converted to reflect similar ranges. As such, the raw score for each child on each subtest was compared to the raw score means provided in the PIPA manual based on the child's age range. This generated a converted score that was the difference between the raw score earned by each child and the mean raw score of the normative sample based on age. The difference between the test mean for the PIPA normative sample and the earned mean scores for the children with clefts as well as the standard deviation for difference between the two are reported in Table 7.

Table 7

Difference between Test Mean for the PIPA Normative Sample and Earned Mean Scores on the PIPA for Children with Clefts

Subtest	Difference	Standard Deviation
Rhyme Awareness	-0.98	3.02
Syllable Segmentation	-0.33	2.95
Alliteration Awareness	-0.80	3.06
Sound Isolation	-0.39	3.46
Sound Segmentation	0.11	2.37
Letter-Sound Knowledge	0.82	7.55

A one-sample t-test was then conducted using the converted scores for each of the PIPA subtests to evaluate whether the mean score earned by children with clefts was significantly different from zero. As reported in Table 8, the sample mean generated by the children with clefts was not significantly different from zero on any PIPA subtest. The results support the conclusion that children with clefts do not have phonological awareness skills that are significantly different than the sample used to generate the norm scores for the PIPA.

While there were no significant between group differences on the PIPA, there were raw score differences between the two groups. As such, the same analysis was performed using the raw data from the children in the comparison group although it was not originally planned. The mean scores for children in the comparison group were converted using the same process described above. These converted scores are reported in Table 9.

Table 8

One Sample Tests for PIPA Subtests for Children with Clefts, test value = 0, df =22

Subtest	t	Significance (2-tailed)	95% confidence interval of the difference	
			Lower	Upper
Rhyme Awareness	-1.56	0.13	-2.29	0.32
Syllable Segmentation	-0.54	0.59	-1.61	0.94
Alliteration Awareness	-1.25	0.22	-2.13	0.53
Sound Isolation	-0.54	0.60	-1.88	1.11
Sound Segmentation	0.22	0.83	-0.92	1.14
Letter-Sound Knowledge	0.52	0.61	-2.44	4.09

Table 9

Difference between Test Mean for the PIPA Normative Sample and Earned Mean Scores on the PIPA for Children without Clefts

Subtest	Mean	Standard deviation
Rhyme Awareness	-1.85	3.23
Syllable Segmentation	-.055	2.45
Alliteration Awareness	-0.74	2.55
Sound Isolation	-0.27	4.57

Sound Segmentation	-0.41	2.59
Letter-Sound Knowledge	0.62	7.76

One sample t-tests were then conducted on the converted PIPA subtest scores to determine whether they differed significantly from zero. As illustrated in Table 10, the sample mean generated by the children without clefts was significantly different from zero on the Rhyme Awareness subtest. These results support the conclusion that the children in the comparison group had rhyme awareness skills that were significantly different, and in fact worse, than the normative sample. The children in the comparison group did not earn scores that were significantly different from the normative scores on any other subtest.

Table 10

One Sample Tests for PIPA Subtests for Children without Clefts, test value = 0, df =22

Subtest	t	Significance (2-tailed)	95% confidence interval of the difference: Lower	Upper
Rhyme Awareness	-2.75	0.01*	-3.25	-0.45
Syllable Segmentation	-1.07	0.29	-1.61	0.51
Alliteration Awareness	-1.39	0.18	-1.84	0.36
Sound Isolation	0.29	0.78	-1.70	2.25
Sound Segmentation	-0.76	0.46	-1.53	0.71
Letter-Sound Knowledge	0.38	0.71	-2.74	3.98

* T-test significant at the.01 level (2-tailed)

The Relationship Between Phonological Awareness and Other Variables

Although no significant differences on measures of phonological awareness were found between the group of children with clefts and the group of children without clefts, the within group relationships between phonological awareness and speech production, language, and hearing status in the group of children with clefts was still of interest given the difficulties the population of children with clefts have in these areas.

Phonological Awareness and Speech Production

Pearson Product Moment Correlation analyses were used to compare standard scores from the GFTA-2 to percentile ranks from each of the PIPA subtests. The mean standard score for the performance of children with clefts on the GFTA-2 was 79 (SD = 21.1, range = 40-118). The results presented in Table 11 demonstrate that only the correlation between standard scores on the GFTA-2 and Alliteration Awareness subtest on the PIPA was significant at the 0.05 level, r(21) = .44, p .035. No other correlations reached statistical significance.

Table 11

Correlations between PIPA and GFTA-2

	GFTA-2 standard score
PIPA—Rhyme Awareness	r = .29; p =.18
PIPA—Syllable Segmentation	r = -.38; p =.078
PIPA—Alliteration Awareness	r = .44; p =.035
PIPA—Sound Isolation	r = .15; p =.49
PIPA—Sound Segmentation	r = .20; p =.37
PIPA—Letter Sound Knowledge	r = .011; p =.96

Phonological Awareness and Language Skills

The sample mean normalized standard scores for the children with clefts on the CELF-P:2 are presented in Table 12. The normalized standard score for the composite scores of the CELF-P:2 have a mean of 100 and a standard deviation of 15 (Wiig, Secord, & Semel, 2004). Pearson product moment correlation coefficients were computed among the CELF-P:2 scores and the PIPA subtests. The results presented in Table 13 show that four of the 25 correlations were statistically significant at the 0.01 or 0.05 level. Receptive Language was significantly correlated with Rhyme Awareness and Alliteration Awareness. Similarly, Language Content was significantly correlated with Rhyme Awareness and Alliteration Awareness. The correlations with other language scales and phonological awareness skills were not statistically significant.

Table 12

Descriptive Statistics for Composite Standard Scores on CELF-P:2 for Children with Clefts

Composite measure	Mean	Range	Standard deviation
Core Language	99.8	77-125	11.9
Receptive Language	97.2	69-115	11.7
Expressive Language	99.4	73-126	12.6
Language Content	99.1	73-115	11.6
Language Structure	98.3	73-121	13.1

Table 13

Correlations between PIPA and CELF-P:2 Composite Scores for Children with Clefts

	Core Language	Receptive Language	Expressive Language	Language Content	Language Structure
PIPA—Rhyme	r =.28	r =.51*	r =.31	r =.56**	r =.27
Awareness	p=.21	p=.02	p=.15	p=.01	p=.22
PIPA—Syllable	r =.13	r =.18	r = -.01	r =.03	r =.07
Segmentation	p=.57	p=.43	p=.95	p=.89	p=.76
PIPA—Alliteration	r =.36	r =.55**	r =.30	r =.64**	r =.26
Awareness	p=.10	p=.01	p=.17	p=<.001	p=.23
PIPA—Sound	r =.24	r =.06	r =.27	r =.07	r =.32
Isolation	p=.29	p=.80	p=.21	p=.75	p=.15
PIPA—Sound	r =.25	r =.01	r =.29	r =.11	r =.23
Segmentation	p=.26	p=.96	p=.18	p=.62	p=.31
PIPA—Letter Sound	r = -.02	r = -.06	r =.04	r =.03	r =.10
Knowledge	p=.94	p=.78	p=.85	p=.90	p=.66

** Correlation significant at the.01 level (2-tailed)

* Correlation significant at the .05 level (2-tailed)

Phonological Awareness and Hearing Status

The impact of hearing status was first explored using the Crosstabs procedure in order to determine if a statistical relationship existed between any of the hearing variables. Passing or failing the hearing screening was not included in the analysis as only two children failed the hearing screening. Three crosstabulations were conducted, examining the relationships between recent ear infections, history of past ear infections, and presence of pressure equalization tubes. In the first crosstabulation, the two variables were a history of recent ear infections within the last two years with four levels (0, 1 to 3, 4 to 6, 7 or more) and presence of PE tubes with two levels (yes or no; see Table 14). While the results of the analysis must be interpreted with caution given the sample size, the Pearson Chi-Squared results revealed that the relationship among these factors was not statistically significant.

Table 14

Crosstabulation between Recent Ear Infections and PE Tubes

Number of recent ear infections		PE Tubes		Total
		Yes	No	
None	Count	6	1	7
	Percent	31.6%	25.0%	30.4%
1-3	Count	11	3	14
	Percent	57.9%	75.0%	60.9%
4-6	Count	1	0	1
	Percent	5.3%	0.0%	4.3%
7 or more	Count	1	0	1
	Percent	5.3%	0.0%	4.3%
Total	Count	19	4	23
	Percent	100%	100%	100%

In the third crosstabulation, the two variables were a history of past ear infections prior to the last two years with three levels (increased, remained consistent, and decreased) and presence of PE tubes with two levels (yes or no; see Table 15). While the results of the analysis must be interpreted with caution given the sample size, the Pearson Chi-Squared results revealed that the relationship among these factors was not statistically significant.

Table 15

Crosstabulation between Past Ear Infections and PE Tubes

	PE tubes		
Past ear infections	Yes	No	Total
Increased	1	1	2
Percent within PE tubes category	5.6%	25.0%	9.1%
Consistent	6	1	7
Percent within PE tubes	33.3%	25.0%	31.8%
Decreased	11	2	13
Percent within PE tubes	61.1%	50.0%	59.1%
Total	18	4	22

In the third crosstabulation, the two variables were a history of past ear infections prior to the last two years with two levels (decrease or no decrease) and history of recent ear infection with two levels (yes or no; see Table 16). The Pearson Chi-Squared results revealed that the relationship among these factors was not statistically significant.

Given the results of the crosstabulations and the small sample size, further analysis using multiple regression examining hearing status as a factor influencing phonological awareness skill level is beyond the scope of this study.

Table 16

Crosstabulation between Past Ear Infections and Recent Ear Infections

	Recent ear infections		
Past Ear Infections	Yes	No	Total
No Decrease	3	6	9
Percent within those with past ear infections	33.3%	66.7%	100%
Percent within those with recent ear infections	50.0%	37.5%	40.9%
Decreased	3	10	13
Percent within past ear infections	23.1%	76.9%	100.0%
Percent within recent ear infections	50.0%	62.5%	59.1%
Total	6	16	22
Percent within past ear infections	27.3%	72.7%	100.0%
Percent within recent ear infections	100.0%	100.0%	100.0%

Next, Kendall's tau correlation coefficients were computed among the individual hearing variables (recent ear infections, past ear infections, and history of PE tubes) and the PIPA subtests (Kendrick, 2000). The results presented in Table 17 show that one of the 18 correlations, the relationship between recent ear infections and performance on the Alliteration Awareness subtest, was statistically significant at the 0.01 level. The positive correlation indicates that children who exhibited recent ear infections tended to perform higher on the alliteration awareness subtest. No other correlations between PIPA subtest scores and individual hearing variables reached statistical significance.

Table 17

Correlations between PIPA and Hearing Variables

	Recent ear infections	Past ear infections	PE tubes
PIPA—Rhyme Awareness	τ =.32	τ = -.24	τ =.37
	p=.12	p=.23	p=.07
PIPA—Syllable Segmentation	τ =.13	τ =.34	τ =.18
	p=.49	p=.09	p=.36
PIPA—Alliteration Awareness	τ =.64**	τ = -.17	τ =.07
	p=.001	p=.41	p=.73
PIPA—Sound Isolation	τ = -.14	τ =.00	τ =.12
	p=.49	p= 1.00	p=.54
PIPA—Sound Segmentation	τ = -.25	τ = -.06	τ = -.03
	p=.20	p=.78	p=.89
PIPA—Letter Sound Knowledge	τ = -.08	τ =.28	τ = -.01
	p=.69	p=.16	p=.97

** Correlation significant at the.001 level (2-tailed)

Phonological Awareness and Oral Reading Fluency

The Oral Reading Fluency (ORF) subtest of the Dynamic Indicators of Basic Early Literacy Skills (DIBELS, Good & Kaminski, 2002) was given to the three children with clefts enrolled in first grade. The results from these three participants cannot be used to draw meaningful conclusions about the group of children with clefts; however, understanding how these three participants performed is informative relative to the other findings of this study.

Given the predictive relationship between phonological awareness and reading abilities in first grade (Mann, 1993), understanding the relationship, as it exists for these three students, will guide decisions about future investigations. Scoring of the oral reading fluency measure takes the median score from the three passages read aloud and assigns a classification, at risk, some risk, and low risk, to the child's performance. It is important to note that this classification takes into consideration the period of the school year in which the assessment was completed, as it would be expected that children would have better skills at the end of first grade as opposed to the beginning of the school year. Of the three children with clefts who completed this measure, one was classified as having some risk, and two were classified as low risk.

Summary of Findings

Overall, there were no differences between children with clefts and children in the comparison group on measures of phonological awareness skills. Additionally, children with clefts did not demonstrate phonological awareness skills that were significantly different than the sample used to generate the norm scores for the PIPA. Children in the comparison group had rhyme awareness skills that were worse than the normative sample, although they did not earn scores that were significantly different from the children with clefts on any other subtest.

Further analyses were conducted to explore the relationships between phonological awareness skills and speech production skills, language skills, and hearing status in the group of children with clefts. Significant correlations were found between speech production and alliteration awareness. Alliteration awareness was also significantly correlated with receptive language skills and language content, as was rhyme awareness. Alliteration awareness and a history of recent ear infections were positively correlated indicating that children with better alliteration awareness also exhibited a greater number of recent ear infections. No other hearing factor was correlated with phonological awareness skills. There were an inadequate number of participants with clefts in the first grade in order to draw meaningful conclusions regarding oral reading fluency skills.

CHAPTER 5

Discussion

Phonological Awareness Skills of Children With Clefts

This study aimed to describe the phonological awareness skills of children with and without clefts. Specifically, it explored phonological awareness skills as they relate to speech production, language development, and hearing status. Based on the sample studied herein, children with clefts demonstrated phonological awareness skills that were similar to their peers. This finding held true in the current study not only when the children with clefts were compared to a matched peer sample, but also when compared to normative sample data that accompanied the measure of phonological awareness that was used in this study.

Although significant differences were not found between the children with clefts and the children without clefts on measures of phonological awareness, follow-up analyses for the group of children with clefts revealed important relationships between phonological awareness skills, speech, and language. Specifically, correlational analyses revealed significant relationships between phonological awareness skills and several different measures of speech and language development. These relationships will be explored in the context of Kamhi and Catts' model of spoken and written language comprehension (1998).

Phonological Awareness and Speech Production

In the current sample of children with clefts, the relationship between speech production and phonological awareness skills was limited in that only one measure of phonological awareness skills was significantly correlated to speech production skills. For children with clefts a moderate positive correlation (r = .44) was observed between performance on the GFTA-2 (Goldman & Fristoe, 2000) and the Alliteration Awareness subtest of the PIPA (Dodd et. al, 2003). This means that children with better speech also performed better on the measure of Alliteration Awareness, while those who had a greater number of speech errors performed less well on the Alliteration Awareness task. One possible explanation for this relationship between speech production and alliteration awareness might be the fact that many of the children with clefts who participated in this study were either currently enrolled in speech therapy or had been enrolled previously. It is possible that their involvement in speech therapy strengthened their alliteration awareness skills. Hesketh, Adams, Nightingale and Hall (2000) demonstrated that children in traditional articulation therapy made gains in speech production and phonological awareness skills, even when phonological awareness was not explicitly taught. While the techniques used by the community based speech-language pathologists who provided intervention for the children with clefts are not known, alliteration awareness skills would be particularly responsive to traditional articulation therapy, as intervention focuses on sound placement within the word, usually beginning with sounds in the initial position. Following this line of reasoning, children who had better speech production skills may have had higher alliteration awareness given their exposure and improvements in speech secondary to therapeutic intervention.

A possible explanation for the lack of relationship between other measures of phonological awareness and speech production may be that children with clefts have adequate phonological representations despite their impairments in speech production. Referring back to Kamhi and Catts' model (1998), it appears that the children with clefts in this sample did not, for the most part, have breakdowns in the auditory analysis or phonological representation phase of auditory processing. It is possible that their speech errors, despite becoming rule based and systematic, remain more closely tied to developmental-type articulation errors as opposed to non-developmental phonological production errors. This distinction could explain why most of the measures of phonological awareness were not significantly correlated to speech production, as their phonological representations remain intact.

Phonological Awareness and Language Skills

In the current sample, several relationships were found between specific language skills and phonological awareness skills. The performance of children with clefts on the Receptive Language composite score of the CELF-P:2 (Wiig et al., 2004) correlated strongly with the Rhyme Awareness ($r = .51$) and Alliteration Awareness ($r = .55$) subtests on the PIPA (Dodd et al., 2003). There are two proposed explanations for these strong correlations. First, a child with lower receptive language skills may have difficulty identifying rhymes and initial sounds in words. This finding appears consistent with the findings of Cooper et al. (2002), which suggested that oral language skills predict phonological awareness skills. Given that no causal relationship can be assumed based on the current study, it is also possible that the participants with poorer performance on the phonological awareness measures have more difficulty learning new words than do other children and therefore have delayed receptive language skills. Whether or not a causal relationship exists and which direction provides the best explanation for the relationship, the current findings do reflect the relationships between phonological awareness and receptive language that are noted in the extant literature.

An alternate explanation for the large correlation between the receptive language composite score on the CELF-P:2 and the Rhyme Awareness and Alliteration Awareness subtests tasks on the PIPA is that the format of these phonological awareness assessment tasks are more similar to the receptive language composite score tasks than are other tasks on the PIPA. The Receptive Language composite score on the CELF-P:2 takes scores from the Sentence Structure subtest, the Concepts and Following Directions subtest, the Basic Concepts subtests (for four year olds) and the Word Classes—Receptive subtest (for five and six year olds). The focus of this composite score, while assessing language comprehension, is also on the assessment of a child's understanding of the form of language structures, including syntactical and morphological concepts. For both the Rhyme Awareness and Alliteration Awareness subtests, children were presented with four spoken words and accompanying pictures and required to identify which item did not belong in the group. The visual supports may have been inadequate to help the children with decreased receptive language skills understand the directions to identify the items that did not belong. Confusion with the instructions, or simply difficulty with the task of identifying the item that did not belong, may have lead to the apparent relationship between these two phonological awareness skills and the child's receptive language score, but not others.

A strong correlation was also noted between the Language Content composite score on the CELF-P:2 and subtests on the PIPA. The Language Content composite was similar to the Receptive Language composite score in that it assessed aspects of language comprehension, but the focus of this subtest is on the meaning behind the various linguistic concepts Strong positive correlations were again found between scores on Language Content and the Rhyme Awareness and Alliteration Awareness subtests (r = .56 and .64 respectively).

The strength of the relationships between these two test items and the Language Content score was even stronger than those noted for Receptive Language Skills. Again, these findings are consistent with the findings of Cooper et. al (2002), that oral language skills predict phonological awareness skills. These findings are also consistent with the findings reported by Rvachew (2006), in that receptive vocabulary was found to have a positive correlation with phonological awareness tasks.

Given the impact hearing status has on language development in the general population, it was important to explore the effects of hearing status on phonological awareness in this sample. The only hearing variable that was correlated to performance on the PIPA was a history of recent ear infections within the last two years. This relationship was most unexpected, since as the number of ear infections a child experienced increased, so did their scores on the measure of Alliteration Awareness. It is suspected that this finding is due to the small sample size and might be nonexistent in a larger study. Alternatively, it is possible that speech-language pathologists follow children who experience more ear infections more closely and for prolonged periods of time perhaps because of the relationship in the literature between language impairments and otitis media. If this were the case, it is possible that children with a history of recent ear infections performed better on the assessments because they have been involved more recently and more intensively in speech language therapy than those children who have not experienced recent ear infections.

Phonological Awareness and Literacy Skills

The study protocol allowed participants to range in age from 4 years 0 months old to 6 years 11 months old, without any specification of the distribution of ages. Only seven 6-year-olds with clefts were enrolled in the study, and of those seven children, only three were in first grade. As a result, oral reading assessments were only given to three of the children with clefts. This is far too small of a sample to draw any meaningful conclusions regarding the oral reading skills of children with clefts.

However, the performance of these three children on the measure of oral reading fluency provides interesting descriptive information regarding reading skills of children with clefts. Based on the median score from the three passages each child read aloud, two of the children were categorized as “low risk” (for subsequent reading difficulties) while the third was classified as “some risk;” none of the three was classified by this measure as “at risk.” The child who was classified as showing “some risk” for reading difficulty demonstrated deficits on the Receptive Language and Language Content composite scores on the CELF-P:2, and also had difficulty on the Rhyme and Alliteration Awareness subtests of the PIPA. This child’s performance was above average for each of the remaining PIPA subtests. Conversely, one of the children classified as “low risk” demonstrated exceptionally high scores on the Receptive Language and Language Content on the composite scores on the CELF-P:2, with relatively high scores on the rhyme and alliteration subtests compared to his performance on other subtests of the PIPA. The remaining child classified as “low risk” performed at a consistently average level on most composite scores of the CELF-P:2 and PIPA subtests. Thus, the child who was identified as having “some risk” for reading difficulties also exhibited deficits on several language and PA measures, while the children identified as “low risk” for reading difficulties demonstrated no such deficits. The performance of these three children taken collectively suggests a trend that supports the relationships found between performance on the language measures and the phonological awareness measures.

The results of this study were consistent with the findings of Richman and Ryan (2003) that suggest phoneme level tasks are a relative strength for children with clefts. As in the existing study, the phonemic awareness skills of children with clefts were indeed a relative strength in that the mean score earned by children with clefts on three of the four phoneme level tasks on the PIPA were above the mean for the normative data. Furthermore, while the mean scores of the children with clefts on phoneme level tasks were slightly above average, the children with clefts earned mean scores on language measures that were slightly below the normative sample values. A larger sample size would again be critical in determining if these differences remain or if it simply is an artifact of this relatively small sample.

Based on the Kamhi and Catts model of spoken and written language comprehension (1998), the children in this sample should ultimately demonstrate strong reading skills. As a group, their mean phonological awareness skills as assessed by the PIPA were well within age expectations. Similarly, their mean scores on the CELF-P:2 measures of receptive and expressive language skills suggest their oral language skills are adequate for sentence and discourse level processing and comprehension. The performance of the three children in first grade who completed the oral reading task provides a small window into the actual reading performance of this group and supports the contention that this group is ultimately at minimal risk for difficulty in learning to read. However, the design of this particular study did not include progress monitoring over time to see if the children ultimately become good readers.

Explanations for Unexpected Findings and Limitations

The results of the current study were not consistent with some of the hypotheses made prior to data collection and analysis. There are four possible explanations for these unexpected findings. First, the high level of achievement attained by the children with clefts on measures of phonological awareness could be explained by their enrollment in speech-language therapy. As previously mentioned, twenty of the twenty-three children with clefts were either currently enrolled or had previously been enrolled in speech therapy at the time of the study. Traditional articulation therapy is commonly advocated as the most effective speech therapy technique to address speech errors in children with clefts (Bzoch, 1997; Golding-Kushner, 2001; Kummer, 2001; Peterson-Falzone, Hardin-Jones, & Karnell, 2001; Peterson-Falzone, Trost-Cardamone, Karnell, & Hardin-Jones, 2006). Traditional articulation therapy techniques include drill based intervention with frequent repetition of target phonemes at the sound, word, and sentence level, so it is not uncommon for a child participating in articulation therapy to be able to report the sound or letter they are working on in speech therapy. It is likely that phonological awareness skills were indirectly addressed using these techniques, and thus such intervention may have been sufficient to increase children's phonological awareness skills for the children in the current sample. In addition, ASHA's current position on the involvement of speech-language pathologists in literacy development (see American Speech-Language-Hearing Association, 2001) emphasizes the speech-language pathologist's role in developing reading skills (Kamhi, Allen, & Catts, 2001; Snow, Scarborough, & Burns, 1999). Given this focus on the role of speech language pathologists in literacy, more and more speech-language pathologists are likely incorporating direct instruction in phonological awareness training in these types of activities. Thus, it is quite possible that the interventions provided by speech-language pathologists acted as a moderator to help develop phonological awareness skills in that group. For these reasons, children with clefts who were in speech-language therapy may have had an advantage over their matched peer group on these measures, as they may already have had instruction, either direct or indirect, in phonological awareness, and they may have already been asked to complete some of the tasks that were included in the assessment battery as part of that experience.

A second explanation for the unexpected findings of the study may be a potential unanticipated selection bias involved with the group of children with clefts. At the beginning of the study, a participant's parent or guardian was required to spend nearly half of a day at Texas Children's Hospital for completion of the testing. This may have excluded potential participants who had parents working full time or those who did not choose to take time off from work in order to participate in the study. This was not the case for the parents of the children in the comparison group. While half of the children in the comparison group were tested at Texas Children's Hospital, the remaining children in the comparison group were tested at their daycare centers. The decision to travel to daycare centers for testing was made when attempts at recruiting typically developing children to come to the hospital for testing did not yield an adequate number of participants. Four daycare centers were used to recruit the remaining participants in the comparison group. Upon visiting several of the daycare centers, the environments varied significantly, as three of the four centers were not "print rich" environments that facilitate literacy skills. In fact, two of the four centers had very little print visible on the walls of the center or books available for the children to read. The children who were recruited from the daycare centers may have had fewer opportunities not only to be exposed to print, but also may have had very different environments to foster their oral language development. As a result, the children with clefts may have had increased opportunities for language and literacy stimulation activities as compared to the children in the study who were cared for at the daycare centers. Future studies will have to control for this experience variable much more tightly.

A third potential explanation for the unexpected findings is related to possible measurement issues. Specifically, the sensitivity of the PIPA is in question given the high percentile ranks children would earn for only correctly responding to one or two items on a particular subtest. For example, a four-year-old child who answered one item correctly on the sound isolation subtest earned a percentile range of 35-39. This percentile range is rather high given that the child produced only one correct answer on the 12-item subtest. However, a child in the same age range who answered two or three items correctly earned a percentile range of 40-44. The difference between these percentile ranges is small, while a child's ability to correctly identify three items correctly as opposed to one item correctly likely demonstrates a sizable difference in whether or not this skill is emerging or the child received credit by chance. This leads one to question the sensitivity of the measurement in separating out those with weak skills and those with adequate phonological awareness skills. In order to examine the possible floor effects of this measure, it is important to examine the raw scores obtained by the children with clefts on the PIPA. Table 18 displays raw scores on the PIPA presented by age group.

By examining the raw scores of children with clefts on the PIPA, it is possible to see that the measure functions well for six-year-olds in that it is able to identify children who are having difficulty with the individual subtests with standard deviations that are less than half the mean score. In contrast, the PIPA appears to function less well for children who are four and five years old with standard deviations that often exceed the mean scores on subtests.

Future research and analysis of the current data set should carefully consider the influence of child age on the individual variables and relationships between the variables in question.

There were also potential limitations with the language measures that were used in this study. Differences in functional language skills are anecdotally reported by speech-language pathologists working with children with clefts and were confirmed by Chapman, Graham, Gooch, and Visconti (1998) in which the conversational styles of children with clefts were different from their peers. The implications of this study are that while standardized measures may not find differences in vocabulary, morphological development, or syntactical skills, children with clefts may continue to demonstrate differences in their use of language.

Table 18

Raw Scores for Children with Clefts on PIPA

	4-year-olds (n = 10)	5-year-olds (n = 4)	6-year-olds (n = 9)
Rhyme Awareness*			
Mean	4.20	5.25	8.56
Standard Deviation	2.86	3.20	3.68
Range	1-10	2-8	2-12
Syllable Segmentation*			
Mean	5.20	6.25	9.22
Standard Deviation	3.74	2.99	2.77
Range	1-11	3-10	4-12
Alliteration Awareness*			
Mean	3.10	2.75	10.11

Standard Deviation	2.89	0.96	3.06
Range	0-10	2-4	3-12
Sound Isolation*			
Mean	3.70	5	11.44
Standard Deviation	4.74	4.55	0.73
Range	0-12	0-11	10-12
Sound Segmentation*			
Mean	0.40	.75	6.78
Standard Deviation	0.70	.96	3.19
Range	0-2	0-2	1-10
Letter Sound Knowledge**			
Mean	6.8	8.25	28.22
Standard Deviation	8.61	8.42	4.24
Range	0-21	0-16	20-32

*Subtest had a maximum score of 12

**Subtest had a maximum score of 32

While both the PIPA and the CELF-P:2 are valuable measurement tools, it is possible that they were not sensitive enough to identify subtle differences across participants.

Finally, consideration was given to whether or not the children in the comparison group were indeed "normal," as they demonstrated phonological awareness skills on certain subtests that were below the performance of the children with clefts. To explore this finding, further attention was given to mean scores for children in the comparison group on measures of language skill and speech production. Table 19 displays the mean values obtained for the comparison group on the GFTA-2 and the CELF-P:2. Clinically, the mean scores obtained for measures of speech production and language development were well within age expectations for the children in the comparison group. While the speech and language skills of the comparison group were certainly within age expectations, their phonological awareness skills were generally weaker than the children with clefts.

Table 19

Statistics for the Comparison Group on GFTA-2 and Composite Standard Scores on CELF-P:2

Measure	Mean	Range	Standard deviation
GFTA-2			
Standard Score	104.5	40-118	11.4
CELF-P:2			
Core Language	101.0	75-127	11.9
Receptive Language	100.0	79-123	11.8
Expressive Language	103.2	75-128	12.9
Language Content	101.7	81-126	12.2
Language Structure	101.5	73-127	12.5

The children with clefts performed far closer to normally paired participants than was anticipated based on multiple studies that have shown that children with significant speech sound disorders are at risk for decreased phonological awareness skills (Bird, Bishop, & Freeman, 1995; Carroll & Snowling, 2004; Larrivee & Catts, 1999; Leitao & Fletcher, 2004;

Sutherland & Gillon, 2005). Additionally, since children with clefts typically perform within the low-average range on measures of language development (Brennan & Cullinan, 1974; Nation, 1970; Pamplona et al., 2000; Philips & Harrison, 1969; Smith & McWilliams, 1968; Spriestersbach et al., 1958), a larger difference between the two groups was anticipated on language assessments. Since the initial sample size estimates were based on expected performance on measures of language skill, and the difference in performance on measures of language skills between the group of children with clefts and the comparison group was much smaller than hypothesized, the number of participants did not meet the minimum that would be required to detect a meaningful difference between groups.

Additionally, the fact that only three children in the group of children with clefts were in first grade and could participate in the oral reading portion of the study limited the ability for the current study to consider oral reading abilities and their relationship with phonological awareness in children with clefts. A larger sample of children in first grade is needed to answer such research questions.

Directions for Future Research

The findings of this study bring up several additional directions for future research. First, continuing this study by increasing the sample size will provide additional information regarding speech, language, and phonological awareness skills in this population. In continuing this study, efforts would be made to eliminate factors that may have created sampling bias, such as providing the option for the participants with clefts to complete testing at the child's daycare centers and elementary schools and recruiting them from those sites. Additionally, larger numbers of participants in first grade would allow for further evaluation of the oral reading skills of children with clefts and it would allow use of multiple regression procedures to consider the amount of variance explained by different variables.

Second, there are multiple directions for follow up studies to this initial investigation. A potential follow up study would include children who had speech disorders without language delay and without clefts as a separate comparison group. The addition of a comparison group (or groups) of children with developmental and non-developmental speech errors would allow further investigation of the impact of speech therapy on phonological awareness skill development, but would also allow further investigation into the nature of the speech impairment in children with clefts. Based on the results of the study presented herein, children with clefts appear to perform like children with speech impairments that are developmental in nature as opposed to those with non-developmental phonological errors. Further exploration of this interpretation is needed. An important feature of any follow up study of this nature would be to include additional tasks that address a broader range of phonological awareness skills similar to those used by Sutherland and Gillon (2005), in order to fully assess skills in Kamhi and Catts' stage of phonological representation (1998).

In addition to studying the impact of the nature of the speech impairment on phonological awareness skills in children with clefts, a longitudinal study would provide information regarding risk factors and outcomes. Therefore, a separate extension of this study would follow the four year olds with clefts who participated in this study through fourth grade. This longitudinal study would provide predictive information regarding the influence of phonological awareness, speech production, and language skills on reading skills in children with clefts, and provide information regarding how those relationships change over time.

Conclusion

This study explored the speech, language, and phonological awareness skills of children with clefts ranging in age from 4 years to 6 years old. Using standardized measures, the performance of 23 children with clefts was compared to a group of matched peers as well as to normative data. Based on this sample, the phonological awareness skills of children with clefts was similar to their matched peer group and to normative data. Further analyses investigating the group of children with clefts revealed a moderate correlation between speech production and alliteration awareness. Additionally, strong correlations were noted between receptive language variables and alliteration awareness and rhyme awareness in the performance of children with clefts; however, no other measure of language skill demonstrated significant relationships with measures of phonological awareness. Relationships between reading skill and measures of phonological awareness and language development could not be assessed in the group of children with clefts secondary to a limited number of participants enrolled in first grade.

Recognizing the impact of oral communication on written language development, this study provides an important foundation for future investigations examining skills that contribute to early reading abilities of children with clefts. Given the positive correlation observed between strong speech production skills and alliteration awareness, it is possible that children with clefts who are participating in speech language therapy to facilitate speech production skills are also receiving training that benefits their development of phonological awareness skills. Additionally, as this study has demonstrated a relationship between weaknesses in receptive language skills and deficits in phonological awareness, it is important for practitioners working with children with clefts to adequately assess both speech production and language skills in order to identify any potential deficits that may interfere with the development of strong reading skills. By understanding the convergence of skills necessary for successful oral and written communication, practitioners can help ensure that children with clefts are equipped for success using both spoken and written communication.

Appendix A:

List of Phonological Awareness Tasks (Gillon, 2004)

Syllable Awareness Tasks

- Syllable Segmentation
- Syllable Completion
- Syllable Identity
- Syllable Deletion

Onset-Rime Awareness Tasks

- Spoken Rhyme Recognition
- Spoken Rhyme Detection
- Spoken Rhyme Generation
- Onset-rime blending

Phoneme Level Tasks

- Alliteration Awareness
- Phoneme Matching
- Phoneme Isolation
- Phoneme Completion
- Phoneme Blending
- Phoneme Deletion
- Phoneme Segmentation
- Phoneme Reversal
- Phoneme Manipulation
- Spoonerisms

Appendix B:

Recruitment Flyers to Parents of Children without Clefts

Speech, Language, and Literacy Skills

Dear Parents,

At Texas Children's Hospital, we are conducting a research study entitled "Speech, Language, and Phonological Awareness Skills in Children with Clefts." The purpose of this study is to determine if children with cleft lip and palate have speech, language, and phonological awareness skills that are different from children without clefts. We are looking for **children without clefts between the ages of 4 and 6 years old, who are developing speech and language skills without difficulty**, to participate in this study.

Parents and children who participate in this research study will be asked to do the following:

- Parents will answer several questions to determine if their child is eligible for the study.
- Parents will complete a brief questionnaire over the phone about the child's medical history (including history of ear infections) and educational history.
- Children will have their hearing screened and complete three speech, language, and phonological tests made for children. These tests should take between one and a half to three hours to complete. In most cases, testing will take place at Texas Children's Hospital.

If you are interested in participating in this study, please contact Kris Wilson at 832-822-3161. Thank you very much for your interest in this research.

Sincerely,

Kristina Wilson, M.A., CCC-SLP
Speech Language Pathologist
Texas Children's Hospital

Appendix C

Demographic Form

DEMOGRAPHIC DATA

Child's Name:____

Date of Birth:____

Year in School:_______________________________________

Parents:____

Address:____

____

What language is spoken at home?__________________________

Does your child have a cleft lip and palate? Yes No

If yes,

What type?________________________________

At what age was your child's lip closure surgery?___________________________

At what age was your child's palatal surgery?______________________________

Is your child: Male Female

Black or African American

Native American and Alaskan

Asian

Hawaiian and Pacific Islander

Hispanic or Latino

White, not Hispanic

Other

What is the highest level of education completed by the child's mother?

Middle School Graduate

Some High School

High School Graduate

Some College

Four-year College Graduate

Graduate School—Masters, PhD, MD, JD

History of Middle Ear Infections:

Over the past 2 years, how many times has your child been treated for otitis media by a doctor/nurse practitioner?

1-3 4-6 7 or more?

How does the number of times your child was treated for otitis media over the past two years compare with the number of times your child was treated for OM in years prior?

- The number of times my child has been treated for OM has *increased* in the last 2 years.
- The number of times my child has been treated for OM in the last 2 years *is consistent* with previous years.
- The number of times my child has been treated for OM has *decreased* in the last 2 years.

Has your child ever had PE tubes in their ears? (PE tubes, also called pressure equalization tubes, are tubes that are surgically placed into the ear drum to help drain fluid from the ear.)

If yes,

At what age were they first placed?

How many sets of PE tubes have been placed?

1 2 3 4 or more

Are the tubes still in?

Both Ears   R ear only   L ear only   Not still in place

Has your child been diagnosed with any of the following:

Cognitive Impairment?	Yes	No
Neurological Impairment?	Yes	No
Syndrome?	Yes	No

<u>History of Special Services:</u>

Speech-language Therapy: Previously enrolled   Currently enrolled   Never enrolled

Describe:

Physical Therapy: Previously enrolled   Currently enrolled   Never enrolled

Describe:

Occupational Therapy: Previously enrolled   Currently enrolled   Never enrolled

Describe:

Special Education: Previously enrolled   Currently enrolled   Never enrolled

Describe:

Other:

School Experience

Is your child in school?____

What grade is your child in school?_____________________________________

Did your child attend one year of preschool?______________________________

Did your child attend two or more years of preschool?_____________________

What is the total number of years your child has been in school?_____________

Appendix D

Lesson Plan Form

Plan:

1. Parental Permission documents

2. Hearing Screening at 35 dB

	500	1k	2k	4k
Right Ear	Pass / fail	Pass / fail	Pass / fail	Pass / fail
Left Ear	Pass / fail	Pass / fail	Pass / fail	Pass / fail

3. CELF-P:2

SNACK

4. PIPA

5. Goldman-Fristoe

Child's Code:_______________________________________

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