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Communication Disorders Associated with Cleft Palate

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INTRODUCTION

Communication

The ability to communicate using speech is a distinctly human characteristic. It is, in fact, what makes us "human" and differentiates us from other species. Communication is a basic human right. It is well known that the presence of a cleft lip and palate or isolated cleft palate may negatively impact a child's ability to communicate effectively and therefore cause significant social, emotional, and educational hardship. Thus, the evaluation and management of communication disorders associated with cleft palate is a critical aspect of comprehensive cleft care.

It is also accepted that children with cleft palate are optimally cared for by an interdisciplinary team of experts working together.[1] Although no single team member can have sufficient breadth and depth of experience across the various disciplines to provide comprehensive cleft care, it is important nevertheless that team members understand and appreciate the contributions of other disciplines and have an adequate appreciation of the subject matter of related disciplines. When this is accomplished, cleft care is ideally transdisciplinary, rather than interdisciplinary or multidisciplinary.[2]

This chapter was therefore developed with the many specialists in mind and not exclusively nor principally for the speech-language pathologist. This chapter emphasizes information about the various aspects of communication that can be affected by clefting in order to facilitate a more complete understanding and exchange between disciplines. The literature and our clinical experience suggest that nonsyndromic cleft lip without cleft palate is not often likely to be associated with significant communication impairment.[3,4] Therefore, for purposes of this chapter, the discussion of communication disorders will focus on children with cleft lip and palate or isolated cleft palate.

When we think of communication disorders associated with cleft palate, we immediately think of speech production abnormalities, especially articulation and resonance. However, it is essential that we understand that communication is made up of several components including hearing, receptive language, expressive language, speech, resonance, voice, and the social use of language most commonly referred to as "pragmatic skills." All these components can be affected by the presence of a cleft. Additionally, a cleft lip and/or palate can also be associated with abnormalities in any one or combination of these communication areas. All these features of communication are mediated by overall cognitive status and a variety of psychosocial variables.

Heterogeneity

A review of the seminal texts and chapters discussing communication impairments associated with cleft lip and palate reveals many areas of agreement and disagreement. It is important to understand when we discuss communication skills in individuals with cleft lip and/or cleft palate this is an expansive topic area. As pointed out by Shprintzen,[5] it is likely that some of the apparent lack of consensus in the literature comes from the fact that this is a very heterogeneous population with one feature in common: the presence of a cleft lip and/or cleft palate. In many cases, the cleft may be an isolated abnormality, or it may be one feature of a multiple malformation syndrome. And so, any discussion of communication disorders associated with cleft lip and/or cleft palate may necessarily be misleading because of generalities. Furthermore, the cleft may be at the source of the communication differences, or it may be associated with a larger constellation of differences of which the cleft is but one feature. As listed in Table 35–1, individuals with cleft lip and palate or isolated cleft palate may present with a number of variables which are known to impact communication.

With respect to discussions of communication, it is of particular importance to differentiate between cleft types. As Shprintzen[5] notes, "Even more fundamental than diagnosing syndromes, clinicians should be aware that clefts of the palate, unilateral clefts of the lip and palate, and bilateral clefts of the lip and palate are not equivalent." The communication disorders associated with each of these cleft types can vary. In summary, there are many variables that impact the presence, type, and severity of communication disorders in individuals with cleft lip and/or cleft palate. In this chapter, we will focus on the communication impairments associated with nonsyndromic cleft palate with or without cleft lip.

Table 35–1.

Variables That May Impact Communication in Individuals with Cleft Lip and/or Palate Thus Contributing to the Heterogeneity of the Population

- Cleft type/severity
- Associated syndromes or other associated conditions
- Age at the time of palate repair
- Efficacy of the palate repair
- Unrepaired residual cleft
- Presence of a palatal fistula
- Status of velopharyngeal function
- Hearing status over time
- Timing, amount and efficacy of communication interventions
- Socioeconomic/linguistic status of the family

A Developmental Perspective

It is well accepted that cleft care is provided over a longitudinal period from birth through late adolescence and that the timing of many dental and surgical protocols are overlaid on physical growth and development. So too should cleft providers be familiar with the developmental course of speech and language. An awareness of the developmental course of normal speech and language and of communication disorders associated with cleft palate adds important information for clinicians as they make treatment plans over the long-term course of cleft care. In this chapter, we will discuss the various components of communication across the developmental continuum. For this purpose, we will divide our discussion of communication into four developmental phases:

- infant (birth to 12 months)
- toddler (12 months to 3 years)
- preschool (3 years to 5 years)
- school age and later

These categorizations correspond with phases of linguistic development that have been utilized widely in the language and developmental literature. They characterize language development in stages that are sequential and distinct from one another. There are two primary phases of communication development. The first is the *prelinguistic* phase, characterized by babbling and gestural communication. The second phase is the *linguistic* phase, characterized by the onset of true words and the development of spoken language. Such a developmental categorization across the different domains of communication is useful for identification of the

aspects of communication that are emerging or of particular importance at a given age and therefore at a given time in the sequence of various forms of physical management. Such a developmental framework is also useful for considerations regarding the types of assessment protocols for evaluation of communication impairments since tests of speech and language development are routinely developed along these lines.

The Relationship between Speech and Language

As pointed out earlier, discussions of the communication characteristics of individuals with cleft palate often focus on speech and velopharyngeal function. It is easy to understand this focus. However, the velopharynx is only one part of a very complex interrelated series of valves that form the human vocal tract. Therefore, even a simple speech screening should take into consideration the structures and processes of the entire vocal tract. Furthermore, speech production and resonance are but one small portion of a larger developmental and communication process. Children with cleft palate, as all children, progress through a sequence or hierarchy of stages that are not necessarily linked to chronological age for many children with congenital anomalies. Speech and language are inextricably linked, especially during early development. This linkage is particularly complex in children with cleft palate who have limitations on oral structure during critical stages of speech and language development. For example, many children with clefts have physical limitations that result in restrictions of early sound systems which can in turn lead directly to reduced early word acquisition.

HEARING

It is well known that middle ear disease and hearing loss are common in children with cleft palate and with many of the syndromes associated with clefting.[6] The topic of hearing and audiologic concerns associated with cleft palate is discussed in detail in other chapters in this book. However, no discussion of communication disorders in children with cleft palate would be complete without reference to hearing. The literature regarding the relationship between hearing, especially otitis media with effusion (OME), and speech and language development is inconsistent. The early literature suggested a significant impact of OME on language development in young, typically developing children without cleft palate. However, a recent meta-analysis of studies on language development and OME shows that the relationship is far less conclusive and more equivocal than previously suggested.[7] Similarly, the current literature regarding the relationship between OME and speech-language development in children with cleft palate does not show a clear one-to-one relationship between middle ear disease and deficits in speech and language performance.[8] Some studies of speech and language development in children with cleft palate have attributed communication delays to OME.[9,10] Other investigations have not shown this same association between hearing and speech and language performance in young children with cleft palate.[11,12] Despite the lack of clear research data showing a direct causal relationship between OME and communication impairment, it is critically important to closely monitor and manage hearing status as part of routine care for children with palatal clefts. Although there may be no evidence of a direct causal relationship between OME and communication impairment in children with cleft palate, it is important to consider that the child with cleft palate has numerous risk factors which are known to negatively impact speech and language development. It is likely that it is this *combination of risk factors* that is of particular concern. The search for causal links between these risk factors and later speech and language development has often revealed complex relationships between such factors, leading researchers to hypothesize a theory of "threshold for impairment." That is, a child may have one severe risk factor (such as speech impairment) or several lesser risk factors (such as OME and mild speech language delay) that push the child over the threshold of impairment. Likewise, the child's risk factors may not exceed the threshold and result in a clinically unidentified impairment. In this model, each risk factor contributes to the child's overall developmental status.[8] Because these risk factors may work together, it is important to address as many of them as possible, including aggressive management of OME.

LANGUAGE

Expressive and Receptive Language

Infant

Most clinicians experienced in the care of children with cleft palate are aware of the potential for impairments in speech production. However, there is a common assumption that there is little importance that occurs in the communication development of affected children prior to the onset of words or prior to palate repair. Anecdotally, this observation is confirmed frequently when medical professionals or families express surprise at the involvement of the speech-language pathologist in the evaluation of babies prior to the onset of speech production. This is a common misconception, one that suggests that communication development during infancy is either nonexistent or of little importance and/or not amenable to evaluation and intervention. To the contrary, however, there are important requisites to later speech and language that develop and that can be affected by the cleft during this early phase.[13]

Until recently, the focus of assessment and treatment of young children with cleft palate has emphasized speech and language problems after they appear. Currently, the focus has shifted from a rehabilitative model that addresses speech and language problems after they are established to a prevention model that addresses problems before they emerge. This recent interest in the speech and language development during

infancy in children with cleft palate stems from the findings of several investigations. Studies of early language development have demonstrated differences in language skills between children with clefts and noncleft comparison groups.[9,14] However, the clinical significance and etiology of these differences is debated. Studies of early expressive language development suggest that children with clefts show delays in the onset and progression of early expressive language development prior to palate repair.[9,14] These findings have been consistent across studies and suggest the importance of the interrelationship between speech and language in early development.

While early vocalizations are the most recognizable milestone within the prelinguistic period prior to the onset of first words, other requisites of speech and language development also emerge during this stage and play a crucial role in establishing the child's interest in communication. Other milestones of early expressive language development include interactional variables, such as turn-taking, and use of gestures for communicative purposes. Although these nonlinguistic variables have not received much attention from researchers, it appears that expression of communicative intent through gestures is a relative strength for children with clefts.[15] However, when early vocalizations that accompany gestures were examined, children with clefts communicated less than noncleft peers.[16]

Another important, though often neglected, aspect of early communication development pertains to the child's understanding of speech. Receptive language development begins during the prelinguistic period and provides a foundation for joint interaction between parent and child that underlies the earliest communicative opportunities for the child. Studies of early receptive language development have shown significant differences between children with clefts and children without clefts; however, the performance of the children with clefts does not fall into a clinically significant range with the exception of children with isolated cleft palate.[9,12,17] This finding suggests a vulnerability in receptive language for some children with clefts at the earliest stages of language development.

The preventive model suggests that, given the importance of the communicative, receptive language, and speech milestones that are emerging during the prelinguistic period, intervention should begin even before palate repair.[13] Borrowing from the early intervention literature, Scherer, D'Antonio, and McGahey[18] explored the use of parent-implemented models of intervention for children with cleft palate. This approach has shown some positive outcomes for early language and speech development while reducing some of the problematic compensatory articulation errors that can persist long after palate repair.

Toddler

The time from 12 to 36 months of age is a critical period for language development. During this time, typically developing children are rapidly expanding their understanding and use of language. For example, the average 2-year-old has an expressive vocabulary of 200–300 words. Further, the size of the child's vocabulary is tied directly to the number of sounds he or she can produce. At this same age, children with cleft palate have a small inventory of consonants that they produce, leading to a cascading effect on their early language development.

Recent studies of early language development in children with cleft lip and/or cleft palate indicate that they show a delay in onset of first words and early expressive vocabulary development. It appears that children with cleft palate often choose words based on their speech sound repertoire, thus leading to limitations in vocabulary development.[9,11,14] These children produce more words beginning with nasals, vowels, and glides and fewer words beginning with oral stop consonants than children without cleft palate.[19] In an intervention study, Scherer[20] found that children with cleft palate learned words with sounds they could produce faster than words with new sounds. Together these studies suggest children with cleft palate display speech sound limitations during the first year of life that impact early vocabulary learning from the onset of first words.

While most studies of language development have focused on expressive language measures and their relationship to speech production abilities, studies of receptive language development indicate that some children with cleft palate experience delays in receptive as well as expressive language. Several studies comparing receptive language development have documented significantly poorer language scores in children with cleft lip and palate when compared to children without clefts.[9,14] However, the clinical significance of the receptive language difference is debated. While group differences reached statistical significance, the scores of the children with cleft lip and palate were often still within the normal range. A recent study[21] compared two groups of toddlers with cleft lip and palate, one with language delays and one without delays. This study showed that the language-delayed group caught up to their peers in receptive language by 3 years of age. While other studies suggest that children with cleft lip and/or cleft palate have language delays that persist into school age, there appears to be a subgroup of children with cleft lip and palate who normalize much earlier. However, these studies also suggest that some children with cleft lip and palate may have a vulnerability in receptive language development that warrants monitoring. As suggested earlier, children with clefts have a variety of risk factors that may combine to impact development. Early receptive language development should be viewed as one of those risk factors that may restrict the progression of speech and expressive language learning and, perhaps, later academic performance.

It appears that some children with cleft lip and palate do show receptive language delays, and it would be beneficial to identify those children early. Recent studies have described a play assessment that may assist in the identification of children with cleft palate who show receptive language delays. A study by Scherer and D'Antonio[22] assessed the symbolic play, language, and speech development of six toddlers between 18

and 30 months of age. They found that performance on the play measure was highly correlated with receptive language development. In a subsequent extension of this study, Snyder and Scherer[23] found that the symbolic play measure successfully predicted the children with cleft lip and palate who had receptive language delays.

While the focus of intervention for children with cleft palate is often speech development, the presence of early language delays suggests that intervention should address both speech and language development. Early intervention methods for young children often dictate a play-based approach for children under 3 years. Scherer[20] found that 2-year-old children with cleft lip and palate improved both vocabulary and speech sound production using a language intervention model. The findings of this study suggest that both language and speech improvement can be achieved simultaneously using language intervention models for young children with clefting.

Preschool

Studies of children with cleft palate continue to show receptive and expressive language differences in the 3–5 year age period when compared to children without cleft palate.[24,25] These deficits do not appear to differ based on cleft type but they do appear to be more associated with those children who demonstrate speech deficits.[25,26] These studies suggest that language impairment persists for some children with cleft palate, particularly for those children with significant speech impairments. Most studies examining language development have identified differences in expressive vocabulary and sentence complexity of children with cleft lip and palate. Scherer[25] found that preschool children with cleft lip and palate had significantly smaller vocabularies and shorter mean length utterances than children without cleft lip and palate. Further, this study determined the size of these group differences to be clinically significant. This recent study supports several older studies that documented language deficits in children with cleft lip and palate.

Some studies suggest that these expressive language deficits resolve by 5 years of age,[27] whereas other studies find that language deficits continue into the school age years. There have been few recent studies examining comprehensive assessment of language functioning in preschool or school age children with clefts.[24,25] Of the few that have provided comprehensive assessment, most include small numbers of participants.[27] While these studies each have identified some areas of language deficit, the results are often at odds with each other. For example, Eliason and Richman[24] found that 4–6-year-old children with cleft lip and palate were delayed in the ability to use verbal rehearsal strategies to mediate verbal problem-solving tasks. However, these same children did not show deficits in more traditional language measures of vocabulary and verbal analogies. On the other hand, Lowe and Scherer[27] showed deficits in some traditional language impairment measures for 5-year-old children with cleft lip palate, such as vocabulary and syntax comprehension. Although these studies do not appear to be in agreement, the studies do point to a persistent language deficit for at least some children with cleft lip and palate. Given the relationship between language performance and school achievement, monitoring of language development throughout the preschool period is essential.

School Age and Later

During the school age period, language impairment may be disguised as an educational impairment. It is not uncommon to see speech and educational testing completed on children while language functioning is never addressed. The strong relationship between language performance and school achievement is well known for children with other disabilities but has not received much attention in children with cleft palate. Several studies suggest that some children with cleft lip and palate, particularly those with isolated cleft palate, continue to show poorer language performance than noncleft peers through school age and into adulthood. However, the extent, characteristics, and persistence of these differences have been debated. Many of the early studies of language performance describe general language delays that include receptive, expressive, and written language modalities extending into adolescence. However, more recent studies suggest that there may be subgroups of children within the cleft population who exhibit different profiles of language performance through school age. One subgroup of children appears to show a general language disability similar to the deficits described in the early studies.[28] These children show deficits that include broad areas of language function (e.g., verbal reasoning, categorization, abstract reasoning, use of verbal mediation for problem solving, rapid naming, and auditory sequential memory).[28,29] This general language disability profile was observed more in males with isolated cleft palate than in children with other cleft types. It should be noted that isolated cleft palate has a higher frequency of association with genetic syndromes and thus puts these children at higher risk of developmental deficits.[30] A second subgroup includes children with expressive language deficits. These children show deficits in rapid naming and auditory memory but not verbal mediation and abstract reasoning. This expressive language group included primarily children with cleft lip and palate.

These two language profiles also exhibit different degrees of risk for academic difficulties.[29,31] The children with general language disability show the greatest risk of reading and math deficits.[32] Whereas the occurrence of reading disability in the noncleft population runs between 10 and 15%, children with clefts show a 30–40% occurrence.[33] A recent study of children with clefts who had been diagnosed with attention deficit disorder found that 50% of those children had learning disabilities, and many of the children had not been previously identified.[32] The association between general language disability and academic difficulties in children with clefts underlines the importance of thorough monitoring of language and academic performance in children with clefts.[34]

Several studies have attempted to identify the source of language and academic difficulties by examining central auditory processing.[35,36] These studies have found differences between children with cleft lip and palate and isolated cleft palate that were detectable at birth and persisted into school age. The studies found poorer temporal processing and discrimination for children with isolated cleft palate than for children with cleft lip and palate and noncleft children. While these measures have been regarded as indicators of language and academic difficulties, the authors suggested that an auditory processing deficit was not the source of the language and academic differences observed in some children with clefts. It is more likely that these impairments are a reflection of the same underlying neural factor.

The persistence of language impairment in some children with cleft palate and the impact of unrecognized impairments on the child's education success indicates that language performance should be assessed thoroughly for those children with poor school performance.

Pragmatics and Social Communication

The literature describing social and pragmatic performance of children with cleft palate has not supported the presence of a pragmatic deficit but has identified aspects of pragmatic functioning that may impact social interaction. It is recognized that children must understand the basic requisites of communicative interaction in order to communicate effectively. These requisites include expressing communicative intent and interpreting social cues of conversational use. While the first requisite appears early in development, the later develops simultaneously with speech and language development. In infancy, children acquire communicative intent, which is the ability to make their communicative needs known through eye gaze, gesture, and/or vocalizations. Children with cleft palate show limitations in use of gestures when combined with vocalizations[15,16] but do not show a deficit in gestural communication alone. This information is important for determining the prognosis for persistent deficits. A child who has no words and demonstrates no communicative intent has a higher risk of slower speech and language development. A child who has no words but who has gestures and is clearly intending to communicate runs less risk of significant speech and language deficits.

During the toddler period, typically developing children begin to overlay speech onto gestural communicative intentions. It appears that children with cleft lip and palate have difficulty with the acquisition of verbal skills and with subsequent use of verbal skills in conversation. Fredrickson, Chapman, and Hardin-Jones[37] examined the communicative functions and conversational structure of children with and without cleft lip and palate. The children with clefts used fewer comments, requests, and disagreements than the children without clefts. In addition, the children with cleft palate were more passive conversational partners in that they used fewer extensions of conversational topics and more topic maintenance than children without clefts. Further, these differences were correlated with articulation performance, suggesting that conversational differences were associated with speech difficulties. In summary, children with nonsyndromic cleft lip and palate or cleft palate do not appear to show specific deficits in pragmatic function but rather a deficit in language use associated with speech intelligibility issues.[38] However, it is important to keep in mind that children with clefts who have associated syndromic conditions may well demonstrate deficits in pragmatic function.

SPEECH

Many texts, chapters, and articles have been written on the speech disorders of individuals with cleft palate. This chapter is not intended to be a substitute for a more detailed review of the literature pertaining to the speech characteristics associated with cleft palate. Rather, in keeping with the themes and goals of this chapter as stated in the introduction, this section is written with the nonspeech pathologist in mind. Additionally, an emphasis is placed on considering speech production as one single part of the broader communication process. Our discussion of the speech patterns associated with cleft palate emphasizes the view that speech is shaped by many dynamic, linguistic processes that are active—and should be appreciated—long before the first words are present in a child's communication process.

A historical review of the literature on speech disorders associated with cleft palate shows a relatively recent move toward greater recognition of a developmental perspective regarding speech impairment. Kuehn and Moller[39] provide an excellent historical review and analysis of the literature on speech disorders associated with palatal clefting. They point out that early characterizations of speech associated with cleft palate emphasized descriptions of the types and frequency of articulation errors compared with normative data. These errors were then often related to anatomic factors such as cleft type, cleft severity, and type and timing of palatoplasty. Gradually, the literature expanded to move from an emphasis on articulation to the description of phonological patterns which acknowledged the higher linguistic processes of speech production. As Moller[40] noted, at this point, the field began to appreciate that much learning was occurring in infants and toddlers prior to age 3 which was the age that speech assessments had traditionally begun. It was acknowledged that the historical and medical propensity for delaying detailed assessments of speech production until the preschool period missed important information about ways in which the child with a cleft was both active and creative in his/her speech acquisition. In more recent years, there has been greater attention paid to how the child with a cleft develops speech sounds and then organizes those sounds into a system that is part of a larger speech and language process. This broader view has taken into account the peripheral and motor aspects of speech production while acknowledging the more central/cognitive aspects of communication. For example, in recent years we have continued to address the articulation

skills of children with cleft palate while attempting to understand their phonological development as well. In this chapter, therefore, we will emphasize speech production as an active, linguistic process with patterns that are emerging from the beginning of sound development and certainly before the onset of first words. While the purpose of this chapter is not to address evaluation methods per se, our discussion of speech and the importance of considering speech production as one part of a more global linguistic/communication process has important implications regarding recommendations for when speech evaluations should be initiated in the clinical management of children with cleft palate and for what types of descriptions are useful for clinical and research purposes.

Articulation and Phonology

In linguistics and speech pathology, *articulation* is a term that describes the physical movements that are involved in shaping the vocal tract above the larynx to produce the various sounds of speech. In most languages consonants are the sounds that carry most of the information that contribute to word meaning. All sounds of any language can be described by the place in the vocal tract where the airstream is constricted and the manner in which the airstream is valved. For example, a "p" sound is made with both lips closed and is therefore called a "bilabial." When producing a "p," the air stream is temporarily held to build up intraoral air pressure and abruptly released. This is called a "stop" because of the stopping of the airstream. Therefore, "p" is a bilabial stop consonant. In contrast, the "f" sound is made with the lower lip touching the teeth, and so it is called a labiodental sound. Here, the air is valved slowly but continuously through the narrowed constriction which is referred to as "fricative." An so, "f" is a labiodental fricative.

Phonology is a branch of linguistics that studies the sound systems of languages. The study of phonology identifies the meaningful patterns of sounds in a language and how these sounds are organized in the mind. Bowen[41] explains that in clinical use, the term "phonology" refers to an individual's sounds system. The gradual process of acquiring adult speech patterns is referred to as "phonological development." Bowen points out that phonological development in children involves three components: the way a sound is stored in the child's mind; the way the sound is actually said by the child; and the rules that connect these two processes.[41]

The distinction between simple description of articulation errors and the more active, linguistically based approach to speech disorders is more than an academic distinction. It has important implications for transdisciplinary issues such as timing of palate repair and measurement of outcome variables. For example, early studies discussed the most beneficial chronological age for palate repair with respect to speech development. A more modern view would emphasize that speech and language age and phonological development are more sensitive and appropriate indicators that should be considered.[42] Similarly, many outcome studies seek to address the impact of a given surgical approach on "speech" by cataloging a variety of articulation errors while not taking into account the child's overall sound system and developmental stage.

One system for describing the speech of young children with cleft palate that can be extremely useful and is worth mentioning is described by Stoel-Gammon and Dunn.[43] This system analyzes a child's speech production capabilities using both "independent" and "relational analyses." A "relational analysis" compares the child's productions with the intended adult model. An error analysis can then examine the type of substitution or omission patterns and can be used to identify the type and frequency of errors compared with the expected adult targets. This analysis system is most similar to that discussed previously in this section whereby early descriptions of the speech of individuals with cleft palate generally were described in relation to normative data. On the other hand, an "independent analysis" documents the child's speech production inventory without comparison to the adult model. This analysis provides information regarding the diversity of sounds and syllable shapes used by children during word attempts. This measure describes the consonants that the child can produce, even though these sounds may be substituted for the correct adult sound. An independent analysis therefore allows for a description of the child's individual articulatory capability and individual sound system. This description of alternative ways of examining the speech production patterns of young children with clefts has meaningful clinical implications. For example, if the child's speech production is compared and contrasted only with the adult model it will surely appear more limited and abnormal than if we assess all of the potential sounds the child can make. It is a common observation that children with cleft palate will come into the clinical speech evaluation with reports describing the child's speech production capabilities in such a way that suggests that the child is "not speaking" or "not saying any words." An independent analysis often reveals that the child has the capability of making more sounds but they may not be using them in word attempts.

Whether conducting a relational or an independent analysis, descriptions of articulation rely primarily on the analysis of the place of articulation and manner of production of sounds and the limitations in these features. However, speech development includes more than learning the physical production of speech sounds. Children acquire a system for organizing sound use that takes into consideration the constraints of the language they are learning. In so doing, children acquire rules that guide their use of speech sounds. In the early stages of speech and language development, when speech motor production is immature, children develop rules for sound use that simplify the motor load on the speech production system. This simplification often leads to omission or substitution of sounds. For children with clefts, substituted sounds may include developmentally earlier sounds or compensatory sounds. Children then establish phonological rules to guide the use of these sounds in words and sentences. Understanding of the phonological rules that children create to

guide their speech production is particularly important when determining intervention goals.

Typically developing children acquire sounds in an ordered manner and progress through well-defined stages of babbling that continue on to meaningful word use.[43] For example, children typically progress from the use of vowels and consonant-like sounds such as "w" or "y" on to the use of true consonants such as "p, t, k" etc. Between 6 and 12 months of age, the number of sounds children use shows rapid increase prior to the onset of first words. By the time typically developing children reach 2 years of age, they are generally using 12–14 different consonants in their speech sound repertoire. For children with cleft palate, the development of these consonants and the consonant sound system is often disturbed in a variety of ways.

Children with cleft palate tend to have the greatest difficulty producing high pressure consonants compared with other classes of sounds. They tend to show a high occurrence of misarticulation for the fricatives and affricates, followed by plosives, glides, and nasals.[8] In the simplest terms, children with cleft palate tend to preserve the manner of articulation while sacrificing the place. In addition to the more common substitution and omission errors, children with cleft palate commonly produce sound substitutions called compensatory articulation errors.[44] The most common and distinctive of the compensatory articulation errors that occur frequently in the speech of individuals with cleft palate is the glottal stop, the result of the child's attempt to move the primary point of articulatory constriction inferior to a malfunctioning velopharyngeal valve. However, it should be noted that compensatory articulation errors are not always a direct result of uncorrected velopharyngeal insufficiency. Such errors may be the result of previous coupling of the oral and nasal cavities that no longer exists. In fact, as pointed out by Hoch et al.,[45] dysfunction of the velopharyngeal valving mechanism can actually be the result of compensatory articulation errors, and improvement in velopharyngeal valving can actually occur through speech therapy aimed at elimination of the compensatory articulation errors. Therefore, the relationship between articulation and velopharyngeal valving is far more complex than is often discussed.[46]

Infant

A number of studies describe the composition of early vocalizations in children with cleft palate.[47,48] During the prelinguistic period, children with cleft lip and palate demonstrate deficits in the onset and composition of their babbling.[49] Their consonant inventories are limited to sounds made by coupling the nasal and oral cavities.[49] Furthermore, the effect of early vocalization deficits appears to persist despite a more normalized speech mechanism following cleft repair. Whereas the vocal limitations of children with cleft lip and/or palate prior to palate repair (at approximately 12 months) do not seem surprising, studies indicate that these limitations often still exist for 1–3 years following repair.[47,48] Additionally, these deficits are apparent regardless of cleft type or early obturation of the palate.[50] Compensatory glottal productions are reported in the vocalizations of children prior to palate repair.[48,51] These glottal productions often take the form of growls or "ohoh" productions and may become embedded into the early sound repertoire of the children with clefts.

Longitudinal studies of the relationship between prelinguistic vocalization and later speech and language performance in children with clefts indicate that children with larger consonant inventories and higher rates of stop consonant production in babbling have better speech and language skills at 3 years of age.[11,52] Chapman et al.[11,52] demonstrated a relationship between small consonant inventories, especially limitations in the production of stop consonants, and later language measures. However, the studies did not find significant correlations between many of the prelinguistic variables and later speech and language measures. Specifically, onset and composition of canonical babbling, which differentiated prelinguistic children with cleft lip and palate from noncleft children, was not significantly correlated with later speech or language measures. While group differences are apparent in the early vocalization patterns of children with and without clefts, there are few clear predictors of later speech and language performance.

Toddler

Following palate repair, children with cleft palate often continue to show speech sound production deficits, including a preference for sounds produced at the extremes of the vocal tract (i.e., labials, velars, and glottals), limited oral stop consonants,[19,49,53] reliance on the phonological processes of backing, nasal assimilations, and use of compensatory errors.[47,48]

These limitations in sound inventory are likely responsible for the early vocabulary deficits observed in children with clefts. Such children produce more words beginning with nasals, vowels, and glides and fewer words beginning with oral stop consonants than children without clefts.[19] Scherer[20] explored the relationship between word learning and speech sound repertoire in an intervention study. She found that young children with cleft lip and palate learned words with sounds that were within their consonant inventories faster than words with sounds that were outside their inventories. Therefore, the children with cleft lip and palate used more words with nasals, glides, and glottals than words with oral consonants.

Longitudinal studies of the relationship between prelinguistic vocalization and later speech and language performance in children with cleft palate indicate that children with larger consonant inventories and higher rates of stop consonant production in babbling have better speech and language skills at 3 years of age.[11,52] Chapman et al.[11,52] demonstrated a relationship between small consonant inventories, especially limitations in the production of stop consonants, and later language measures. However, the studies did not find significant correlations between many of the prelinguistic variables and later speech and language measures. Specifically, onset

and composition of canonical babbling, which differentiated children with cleft palate from children without clefts in the prelinguistic period, was not significantly correlated with later speech or language measures. Although group differences are apparent between the early vocalization patterns of children with and without clefts, there are few clear predictors of later speech and language performance.

Preschool

Children in the 3–5 year age range continue to exhibit speech impairments that are characterized by developmental errors, nasal substitutions, compensatory articulation, and persistence of phonological process errors. Hardin-Jones and Jones[54] examined the speech of 212 preschool and school-aged children with cleft lip and palate. Approximately 13% of the children used nasal substitutions and 25% used compensatory articulation errors, specifically glottal stop substitutions. The findings indicate that these error patterns persist for a substantial number of children despite the fact that 68% of the children in this study had received speech therapy.

Chapman[55] examined the use of phonological processes in 3–5-year-old children with and without cleft lip and palate. This study showed that children with cleft lip and palate use phonological processes for a protracted period of time. Phonological processes are a set of rules the children use to simplify speech production during early language development.[43] For example, young children may omit the final consonant in words (e.g., "ba" for ball) or omit the first syllable of a word (e.g., "jamas" for pajamas) in order to ease the production task. However, children also substitute sounds in predictable ways such as substituting sounds made in the back of the mouth for sounds in the front of the mouth (e.g., "gagi" for daddy). Chapman found that young children with cleft lip and/or palate used these simplification rules longer and used some processes more than children without clefts. Significant differences were found between the groups at 3 and 4 years but not by 5 years of age. The predominant processes that differentiated the groups at 3 years of age include backing (e.g., "kea" for tea), stopping (e.g., "do" for zoo), stridency deletion (e.g., "un" for sun), final consonant deletion (e.g., "ba" for ball), syllable reduction (e.g., "jammys" for pajamas), and cluster simplification (e.g., "top" for stop). The authors suggest that the first three processes may be associated with difficulty producing high pressure consonants since these processes include sounds in the stop and fricative manner categories that are problematic for children with clefts. The remaining processes are observed in the speech of children with phonological and language impairments without clefts and may result from the children's attempts to reduce speech production complexity given a limited consonant inventory.

During the preschool phase, the child's sound inventory begins to approach the adult model. Many of the developmental phonological processes are typically eliminated by this age. When speech sound errors persist, this is the age at which direct speech therapy is most likely to begin. During the preschool period the primary articulation patterns that characterize children with cleft palate include limited phonetic inventories, poor speech accuracy, and presence of compensatory articulation errors. Scherer[56] described the speech of 25 children with clefts and 25 children without clefts between 3 and 5 years of age. The children with clefts showed significantly poorer articulation scores and speech accuracy[57] than the children without clefts. A severity index associated with the speech accuracy score placed the children in the mild to moderately impaired range, an improvement over the toddler period. Although the children's speech remained behind those of age matched peers, children with clefts were making advances in speech production. The most notable category was the fricative category (e.g., /f/,/v/,/th/,/s/,/z/,/sh/,/3/). Percent correct use of this category improved from 38% to 67% from 3 to 5 years.

The persistence of compensatory errors during the preschool years often provokes the onset of speech therapy for many children with clefts. Studies have suggested that approximately 25% of children with clefts use compensatory articulation errors, and glottal stops appear to be the predominant error pattern.[51,54] For children who use compensatory articulation patterns to a significant degree, speech intelligibility may be severely reduced. Further, when this pattern becomes habituated during the preschool years, it can be particularly resistant to change in therapy.[58,59]

As discussed earlier, in addition to phonetic features of speech associated with the physiologic aspects of the cleft, children with clefts show difficulty with the acquisition of the rules of the sound use. Phonological processes, or the rules used to simplify classes of sounds, are typically eliminated during the preschool period. Chapman[55] examined the use of phonological processes in children with clefts. She found that children with clefts used phonological processes for a longer period of time than children without clefts, but by 5 years of age these children had caught up to their peers. The processes that distinguished the children with clefts included processes that omitted sounds or syllables, such as final consonant deletion (e.g., "ba" for ball), syllable reduction (e.g., "jammys" for pajamas), stridency deletion (e.g., "hou" for house), or cluster simplification (e.g., "top" for stop) and processes that substituted compensatory sounds or developmentally easier sounds, such as backing (e.g., "kov" for stove), glottal replacement (e.g., "cu?" for cup), stopping (e.g., "knip" for knife), and deaffrication (e.g., "shicken" for chicken). While some of the substitution processes are more often associated with high pressure consonants that are problematic for children with clefts, the omission processes are observed frequently in the speech of other impaired groups of children, such as those with phonological and language impairments.

School Age and Later

Children with clefts continue to make progress in their speech development during the school-age years, although this progress is more rapid for the younger children than

older children. As adults, individuals with clefts still are consistently judged to have poorer speech than individuals without clefts.[47,60,61] Bardach et al.[61] reported that articulation was judged to be within normal limits for 57% of adolescents with bilateral cleft and palate. This figure is similar to the 55% reported by Peterson-Falzone[62] for adolescents with a variety of cleft types. While progress is evident during the school years, a substantial number of children still do not attain normal speech production by adulthood.

The presence of unresolved compensatory articulation errors is a major concern for a subgroup of children with clefts during the school age years. Although this subgroup may be small in number, they represent a particularly challenging group of children for the speech-language pathologist. In most cases, velopharyngeal dysfunction will be addressed prior to or within the first few years of school; however, the impact of years of compensatory articulation use is often not readily overcome.[63] Further, some studies exploring the relationship between speech and language performance suggest that those children with poor speech intelligibility due to compensatory articulation errors often also have significant language impairment.[21] These findings suggest that the articulation/phonological impairments, may affect the organization of other related developmental parameters, including language and reading.

Dentition

The impact of dentition on speech production skills of children with cleft palate is a testimony to the human speech production mechanism to adapt. As we know, oral clefting can be associated with a wide range of dental and occlusal abnormalities. An excellent review of the dental/occlusal problems that can impact speech production is summarized by Peterson-Falzone, Hardin-Jones and Karnell.[8] They review the potential impact of a protrusive premaxilla, retrusive premaxilla, crossbite, low palatal vault/open bite, missing teeth, rotated anterior teeth, and ectopic teeth on articulation. While any or all of these dental abnormalities can impact speech, particularly articulation, there are few reports that show clear causal links between these features and speech articulation. Rather, the speech production system appears imminently capable of aerodynamic and acoustic compensations for most dental abnormalities.

Resonance and Velopharyngeal Function

The communication disorders most commonly associated with individuals with cleft palate are those related to velopharyngeal dysfunction or incomplete separation of the oral and nasal cavities. When this separation is disturbed, a variety of alterations in speech can occur, including hypernasality, mixed resonance, cul-de-sac resonance, weak pressure consonants, and compensatory articulation patterns. The strong relationship between palatal clefting and the presence of hypernasality is so well recognized that in the early years of the field, the presence of hypernasality in a speaker was commonly referred to as "cleft palate speech." However, it is important to know that not all individuals, nor even most individuals, with cleft palate will demonstrate hypernasality or other speech symptoms associated with faulty separation of the oral and nasal cavities. Conversely, hypernasality and symptoms of velopharyngeal dysfunction may or may not signal the presence of a cleft or submucous cleft palate.

Definitions of several of the symptoms associated with abnormal velopharyngeal valving are presented in Table 35–2. However, it is worth reiterating some important distinctions regarding some of these speech characteristics. Hypernasality is a *resonance disorder*. It refers to inordinately high nasal resonance on vowels and vocalic consonants. It is the result of abnormal coupling of the oral and nasal cavities and is a physical phenomenon that is typically assessed by perceptual means. There is wide variability across languages (and even across dialects of a given language) in terms of what is normal or acceptable nasal resonance. On the other hand, nasal emission is an *articulation disorder*. Most simply defined, it is the passage of air through the nose for high pressure consonants that should not be associated with any nasal airflow. Nasal emission may be audible or inaudible. It is often associated with reduced intra oral air pressure. Hypernasality and nasal emission may occur in the same speaker and often do coexist, but they are not the same phenomena. Additionally, both these features are the result of *supraglottal* disturbances and therefore should not be referred to as *voice disorders*, a term reserved for disorders of the larynx and phonation.

A listener does not require specialized training to hear many of the effects of abnormal oral nasal coupling on a speaker's speech quality and intelligibility. While the untrained listener might be equipped to identify the presence of a speech abnormality, it is a far more difficult and complex task to identify the cause(s) of that disorder and to develop an appropriate and effective management plan.

In particular, the role of the velopharynx in speech production and the nomenclature used to describe disturbances in this system are far more involved than is sometimes acknowledged in discussions of communication disorders associated with cleft palate. In its most simple form, the speech production process can be thought of as a large air-filled container always closed at the bottom with two openings to the atmosphere, the lips, and the nares. In this container, there are several valves that can be opened or closed to varying degrees, thus changing the shape of the container and the resistance to airflow. These valves include the larynx, the velopharynx, the tongue and lips, and the nasal passages. Airflow must move through this series of valves in a coordinated and tightly timed manner, thus creating a series of rapidly changing air pressures and airflows that we ultimately perceive as the sounds of speech. Viewed most simplistically, the role of the velopharyngeal valve is to separate the oral and nasal cavities during speech and swallowing. For speech, the velopharynx directs air from the lungs and larynx through the mouth for oral sounds and through the nose for nasal sounds. When this valving is disturbed, speech can be affected in several ways, as described above.

Table 35–2.

Resonance, Articulation, and Phonation Disorders Frequently Associated with Cleft Palate and/or Velopharyngeal Dysfunction

Hypernasality The perception of inordinate nasal resonance during the production of *vowels*. This results from inappropriate coupling of the oral and nasal cavities. (The term *inordinate* is used because low vowels and vowels in nasal consonant contexts are normally somewhat nasalized).
Nasal emission Nasal air escape associated with production of *consonants* requiring high oral pressure. It occurs when air is forced through an incompletely closed velopharyngeal port or a patent-oral nasal fistula. Nasal emission may be audible or not. *Note: Hypernasality and nasal emission* are not synonymous, although they often occur together and are both symptoms of velopharyngeal dysfunction.
Hyponasality A reduction in normal nasal resonance usually resulting from blockage or partial blockage of the nasal airway by any number of causes, including upper respiratory tract infection, hypertrophied turbinates, and a wide, obstructing pharyngeal flap.
Hyper-hyponasality (mixed resonance) The simultaneous occurrence of hypernasality and hyponasality in the same speaker usually as the result of incomplete velopharyngeal closure in the presence of high nasal cavity resistance that is not sufficient to block nasal resonance completely.
Cul-de-sac resonance A variation of hyponasality usually associated with tight anterior nasal constriction often resulting in a muffled quality.
Nasal substitution The articulators are placed appropriately for an intended oral consonant. However, incomplete velopharyngeal closure causes the sound to be produced as a nasal consonant. For example, b becomes m and d becomes n. Such substitutions frequently are called homorganic nasals.
Compensatory articulation The articulators are placed inappropriately so as to enable creation of the plosive or fricative characteristics of the sounds they replace. For example, if a patient cannot build up oral pressure for the fricatives (e.g., s) or plosives (e.g., p) because of velopharyngeal dysfunction, they may create those pressures below the level of the velopharyngeal port. Such substitutions include glottal stops, pharyngeal stops, and pharyngeal fricatives among others.
Sibilant distortion Inappropriate tongue placement for the sounds /s/ and /z/.
Laryngeal/voice symptoms A variety of phonation disorders may accompany velopharyngeal dysfunction, including hoarseness, low speaking volume, strained or strangled voice quality, and unusual pitch alternations. One theory for the co-occurrence of velopharyngeal and laryngeal symptoms is that speakers with velopharyngeal dysfunction may attempt to compensate for the inability to achieve complete closure and maintain adequate speech pressures by compensatory activity at the level of the larynx.

Modified from D'Antonio L, Scherer NJ. The evaluation of speech disorders associated with clefting. In Shprintzen RJ, Bardach J (eds). Cleft Palate Speech Management, St. Louis: Mosby Elsevier, 1995, pp. 176–220.

Historically, the velopharynx has been viewed as a simple binary valve with two positions: open or closed. However, research and clinical observations have shown that the velopharynx is a complex three-dimensional valve with a variety of shapes and patterns of activity that differ among speakers.[64] Just as the oral articulators, such as the lips and tongue, have varying degrees of shapes and movement patterns, so too does the velopharynx assume different shapes and positions for different sounds. In addition, it is not enough that the velopharyngeal valve be capable of achieving complete closure; it must do so in a tightly controlled time domain in coordination with other articulators.

As discussed earlier in this chapter, the child with a cleft, like other children, is engaged in an active process of learning the speech sounds and patterns of the ambient language. Disturbances in anatomy during early speech and language development can interact with more global linguistic processes that can impact the speech sound system of a child. Therefore,

it is important for the various professionals involved in the care of children with cleft palate to be aware that the presence of symptoms of velopharyngeal valving disorders may have a variety of compound and sometimes inter-related causes that have their roots in current anatomic limitations or in earlier learning in the presence of abnormal anatomy. It is thus essential that velopharyngeal valving be appreciated with this understanding of learning and higher-level linguistic processes in mind.

Terminology

Traditionally, when symptoms were present that suggested the velopharynx was not functioning correctly, such was referred to as "velopharyngeal incompetence" or "VPI." However, hearing hypernasality or nasal emission (i.e., speech symptoms associated with VPI) does not necessarily indicate that the velopharynx cannot achieve closure. Rather, it simply means that in this instance it did not achieve closure. Such misunderstanding of the complexity of velopharyngeal function can have profound diagnostic implications. For example, the diagnosis of VPI suggests that the velopharyngeal mechanism cannot achieve closure, and for many cleft care providers it suggests that only physical management will correct the problem. In addition, this label will often bias the unfamiliar speech pathologist to believe that additional speech therapy is not warranted until physical management is completed. Therefore, it is important that our language, regarding the causes of the symptoms associated with abnormal nasal resonance and velopharyngeal valving abnormalities, be precise and based on the underlying physiology. There have been many discussions in the literature regarding the most appropriate language to use in describing velopharyngeal valving disorders.[46] There appears to be a trend in the more recent literature to use the term "VPI" (velopharyngeal incompetence or velopharyngeal insufficiency) when diagnostic studies have clearly determined a true physical limitation, whereas "velopharyngeal dysfunction" is used when it is clear that there is some malfunction, but the cause of such remains unclear.

Differential Diagnosis

There are a variety of factors that can result in speech symptoms such as hypernasality or nasal emission, i.e. symptoms most often associated with velopharyngeal dysfunction. Some of these are structural and include a true inability of the velopharyngeal port to achieve closure due to an absence of adequate tissue to allow for closure; improper muscle insertion of the levators preventing closure due to poor motion or a residual central trough in the nasal surface of the palate; the presence of an oral nasal fistula; interference from other structures such as the tonsils, adenoids, and nasal passages; or neurologic impairment. There are also a number of speech-related variables that can impact the presence of velopharyngeal symptoms, including articulation and phonologic patterns, inconsistency of speech patterns, the role of phonetic context, rate and timing of speech, and fatigue. The role of the speech-language pathologist in evaluating the speech of children with cleft palate is to determine which symptoms are the result of true physical limitations (and which therefore require physical management) and which symptoms are more related to learning or habituation of patterns (and which therefore require behavioral intervention, i.e. speech therapy).

This process of differential diagnosis can be quite difficult since speech symptoms that seem to be similar and, in fact, indistinct from one another to the casual listener may be varied in cause and, therefore, in appropriate management. For example, in patients with cleft palate, hypernasality and nasal emission may occur in the presence of a repaired cleft and a residual oronasal fistula. Casual perceptual observations may not reveal the source of these symptoms. In fact, in our experience it has been common for some team members to refer to the speech symptoms associated with a fistula as "VPI." However, it would be inappropriate to label the phenomenon as VPI without investigation. In some instances, the symptoms may be solely attributable to air escape through the fistula. In this case, the diagnosis would be hypernasality and nasal emission caused by a patent oronasal fistula, and the symptoms may thus be completely unrelated to the velopharyngeal mechanism in any way. The appropriate management would be repair or obturation of the fistula. In other cases, the symptoms may appear to be attributable to a lack of proper velopharyngeal function or to a combination of causes. For example, there are reports in the literature that have shown a relationship between temporary obturation of a patent oronasal fistula and associated improvement in velopharyngeal valving.[46,65–67] Another illustration of the need for differential diagnosis is the phenomenon of phoneme specific-VPI.[68] This is the presence of nasal emission that is isolated to specific pressure consonants, most commonly the fricatives /s/ and /z/. The inexperienced clinician often mistakes this rule-based phonological error as a sign of true VPI. Many patients with sound-specific nasal emission such as this are referred for surgical management for what is actually an articulation/phonological error pattern.

These examples emphasize the point that the resonance and velopharyngeal valving disturbances associated with cleft palate are often complex and multifactorial and require in-depth evaluation to sort out the variety of potential etiologies. Additionally, the amount of information necessary for making these differential diagnoses varies over the child's developmental course and interacts with chronologic age, developmental age, language stage, and speech development profile. In many cases, therefore, the diagnostic process must be conducted over time as the child continues to develop and in relation to other surgical, dental, and behavioral interventions.

Infant and Toddler

In the early stages of communication development, there are few insights into whether velopharyngeal valving will be adequate for speech or not. It is not possible to judge velopharyngeal function directly in these early stages of development.

However, there are some indirect links or indicators that are worth watching carefully, including phoneme repertoire and the emergence of compensatory articulation patterns, particularly the glottal stop. At this age, there is not enough speech and language development to make clear determinations regarding whether or not the velopharyngeal port is functioning or will function adequately for normal speech production.

Some young children, especially in the toddler and early preschool age range, may demonstrate inconsistent nasal emission that appears to be related more to a failure to have achieved the correct manner distinction between oral and nasal sounds. This error pattern is more related to a phonological process than to true VPI. That is, the child may not have acquired an understanding of the difference between oral and nasal sounds. In many instances, the very young child or the child with a speech and language delay may not discriminate between oral and nasal contrasts in his or her own productions or phonological system or in the models of the speech pathologist. Many young children who present with inconsistent nasal emission are stimulable for correct production of oral consonants once they recognize and understand the difference between oral and nasal airflow. It is therefore important to differentiate in such children between nasal emission due to an incompetent VP valve and nasal emission that is the result of an articulation/phonological based pattern.

Preschool

During the preschool years, resonance and velopharyngeal function can be assessed with some accuracy. As the child's sound repertoire, articulation skills, and expressive language expand, there are more opportunities to evaluate whether velopharyngeal valving is adequate for speech production. Generally, the more developed the child's speech production skills, the more accurately velopharyngeal function can be assessed. It is often difficult during the early preschool phase to sort out how much of the perceived velopharyngeal symptoms are the result of habituated patterns or phonological processes and how much are the result of true velopharyngeal insufficiency. However, careful evaluation and monitoring can result in accurate differential diagnosis.

When there are severe and consistent velopharyngeal symptoms, it is typically easy to make a determination regarding the diagnosis and therefore make appropriate treatment recommendations. Even after thorough evaluation, however, it can be very difficult in some cases to determine whether or not the velopharyngeal mechanism is adequate for speech production. In these cases of borderline or variable velopharyngeal function (or when performance appears inconsistent), it is difficult to determine whether or not the velopharyngeal mechanism is adequate for speech and therefore whether surgical management or speech therapy is the most appropriate treatment recommendation. In these complex cases, it is useful to use a circumscribed period of speech therapy to provide additional and often necessary diagnostic information.

Typically, it is during the preschool period that such diagnostic therapy becomes possible. When velopharyngeal function is variable (as it often is in young children), it is useful to attempt stimulability testing.[46,69] A cornerstone of modern speech therapy is the belief that a child's ability to be stimulated for improved speech production through auditory, visual, and in some instances tactile models and cues is a good prognostic indicator of the potential for long-term improvement. Such stimulability testing is particularly useful in the child with variable velopharyngeal function. It can provide valuable information about whether behavioral management is likely to remediate velopharyngeal symptoms or if it appears that physical management is indicated.

Morris[70] suggests that there are two major subgroups of children with marginal velopharyngeal dysfunction and that they can be most easily distinguished by their response to short-term therapeutic intervention. The first is the "almost-but-not-quite" (ABNQ) subgroup. This group tends to present with mild consistent nasalization of speech that is highly consistent among and within tasks. Morris suggests that speech therapy is not likely to be successful with this group. A brief period of therapy for the young child with inconsistent VP function should reveal whether further improvement is possible.

The second diagnostic group of marginal velopharyngeal function described by Morris is the "sometimes-but-not-always" group (SBNA). Children in this group generally show marked inconsistency in velopharyngeal function. Some children in this group will show improvement in VP function with training and some will not. It is essential to consider the children in this group who do not improve. A careful analysis of their speech errors is necessary to determine if the errors are random or if they appear to be rule-governed and part of a more encompassing phonological system. For example, it is common to see preschool children with cleft palate who can produce all or most of the stop consonants with no inappropriate nasal airflow. However, often all or most fricative and sibilant consonants are nasally emitted. In these cases it is often true that the child has developed a system where friction consonants are marked by nasal emission or substituted by a true compensatory articulation error, in particularly the posterior nasal fricative, and a thorough description of the error patterns will assist in the diagnostic process. Furthermore, a brief period of speech therapy will also provide invaluable information concerning whether velopharyngeal closure can be facilitated for the incorrectly produced sounds.

It is these children with inconsistent velopharyngeal function who present the greatest dilemma for the surgeon and speech pathologist. Especially in young preschool children, the inability to achieve consistent velopharyngeal closure is commonly related to several contributing variables. Because of the multiple, interrelated variables that can impact velopharyngeal function in these children, it is very important that decisions to provide surgical alternatives be provided only after thorough evaluation and counseling. In many instances, it is these patients who have poor surgical outcomes

since the original problem was, in fact, multifactorial. It is these patients with "diagnostic dilemmas" who require intensive counseling prior to surgical intervention in order to facilitate realistic expectations for postsurgical outcomes.

It is also during the preschool stage that questions regarding whether or not a palatal fistula is symptomatic can be addressed. We know that hypernasality is the result of abnormal oral nasal coupling. As discussed above, audible nasal emission may be attributable to inadequacy of the port or to articulatory errors. However, as mentioned previously, both symptoms may also result from an oronasal fistula with or without VPI. Significant controversy exists in the literature concerning accurate identification of symptomatic fistulae, the extent of the effects of such on speech, and decisions regarding surgical repair. There are many opinions expressed in the surgical literature, and there is a common misconception that there is a relationship between the size and location of a palatal fistula and its effects on speech. A more conservative view is that the functional significance of a palatal fistula on speech must be determined for each patient individually. D'Antonio et al.[71] showed a significant improvement in perceptual judgments of hypernasality, frequency of nasal emission, perceived oral pressure, and speech quality when comparing speech ratings with fistulae unoccluded and temporarily occluded with chewing gum. However, there was no consistent relationship between the improvement in speech characteristics between the unoccluded and occluded conditions based on size or location of the fistulae. Results of the same study suggested that an important factor influencing changes in speech and aerodynamic characteristic when a palatal fistula is obturated is the individual's nasal cavity resistance. Additionally, Isberg and Henningsson[65] showed a relationship between temporary obturation of a patent oronasal fistula and concomitant improvement in velopharyngeal valving. Results from these studies concerning palatal fistuale are presented here to emphasize that a number of factors are likely to contribute to the effects of palatal fistulae on speech. Therefore, when a palatal fistula is observed, statements concerning its effect on hypernasality, nasal emission, or speech quality should be made with great caution. For many children the process of determining whether the fistula is symptomatic for speech or not begins in the preschool age range but is difficult to determine before this age when the child is capable of cooperation.

In addition to true VPI articulation and phonological-based errors, and the contribution of palatal fistulae, there is one other important yet infrequently discussed potential cause of hypernasality and nasal emission that for some clinicians is counterintuitive. In some children, hypertrophic tonsils can prevent complete VP closure, thus resulting in hypernasality and nasal emission or both.[72,73] It should be noted, however, that hypertrophic tonsils and adenoids may also impact speech by blocking the flow of air through the velopharyngeal port and may thereby also cause hyponasality or denasality. The decision to remove tonsils in a patient with a cleft palate should therefore be accompanied by a thorough perceptual and instrumental speech evaluation.

As this discussion of tonsils and adenoids suggests, and as shown in Table 35–2, individuals with cleft palate may also demonstrate hyponasal resonance, mixed hyper-hyponasal resonance, or cul-de-sac resonance. Although hypertrophic tonsils and adenoids are the most common cause of these resonance abnormalities, other factors can result in these symptoms. Patients with repaired palatal clefts frequently have structural deviations of the nasal airway that can result in a resistance to nasal airflow. Additionally, children with cleft palate are subject to the same sources of anterior nasal airway obstruction as noncleft patients, such as allergic rhinitis and other nasal airway changes. Warren[74] and Dalston and Warren[75] have suggested that the nasal cavity is an important factor affecting not only resonance, but articulation and velopharyngeal function as well. Therefore, when hyponasality is present, such should be monitored on an ongoing basis. If the symptoms persist or are reported to be chronic, then further evaluation is indicated, and referral for otolaryngologic evaluation should be considered.

School Age and Later

When velopharyngeal symptoms persist into the school age years, it is critical to establish an aggressive monitoring or management plan to normalize the child's speech as soon as possible in order to avoid social and psychological concerns. Some children continue to present with VPI into the school age years. There is significant variability in treatment protocols across centers, and with a mobile population and follow up difficulties some children may present to a new team or after a hiatus in care. They may therefore present with significant velopharyngeal symptoms that have gone undiagnosed, untreated, or incompletely treated.

The most notable risk for deterioration in velopharyngeal function during the school age years and later relates to changes in the dimensions of the pharyngeal cavity caused either by adenoid involution or maxillary advancement. As the adenoids involute, some children with cleft palate may experience difficulty achieving complete velopharyngeal closure, as the palate can no longer stretch to accommodate the increasing distance of the pharyngeal depth.[76,77]

Similarly, in patients who require advancement of the maxilla through either surgical advancement or distraction, there is a risk that movement of the maxilla and therefore the palate will result in VPI.[78–81] Following either procedure, patients may experience a temporary period of hypernasality or nasal emission. In some instances, surgery may result in permanent velopharyngeal symptoms that will require physical management.

Voice and Phonation

It is a common clinical observation that children with cleft palate have a high occurrence of voice symptoms such as hoarseness, breathiness, low intensity, and abnormal pitch.[82,83] Additionally, it has been shown that children with cleft palate demonstrate a high occurrence of laryngeal pathology including vocal nodules, vocal fold thickening,

edema/inflammation, incomplete glottal closure, and hyperconstriction.[83] Although the prevalence of voice disorders in this population is unclear, the data suggest that phonation disorders are more frequent in children with cleft palate than children without clefts. The relationship between cleft palate and laryngeal dysfunction may be congenital or behavioral. For example, as mentioned previously, palatal clefting may be associated with a number of multiple malformation syndromes for which resonance and phonation disorders frequently co-occur. Also, as we have discussed previously, the vocal tract is a complex series of interrelated valves. Impairment in one valve may lead to compensatory activity or impairment in another. Specifically, speakers with impaired velopharyngeal valving may use increased respiratory effort or abnormal laryngeal valving, both of which are potentially damaging to the larynx and which may result in observable laryngeal pathology, voice symptoms, or both. D'Antonio et al.[83] reported that 41% of 85 patients with symptoms of velopharyngeal dysfunction had abnormal voice characteristics and/or observable laryngeal abnormalities. McWilliams et al.[82] reported on 32 children who were described as chronically hoarse who underwent instrumental assessment. Eighty-four percent of these children had some vocal fold pathology, and 59% had borderline velopharyngeal valving. McWilliams et al.[82] reported that alteration of velopharyngeal valving in children with velopharyngeal valving disturbances and laryngeal pathology demonstrated improvement in voice symptoms following management of velopharyngeal symptoms. These data suggest a relationship between velopharyngeal valving and laryngeal pathology. The authors further suggest that some children compensate for minimal VPI by increased laryngeal valving.

Regardless of the cause of the relationship between clefting and phonatory/laryngeal abnormalities, it is important that laryngeal/voice function be screened routinely in children with craniofacial anomalies. For infants, rapid development of laryngeal/voice function takes place in the first 12 months of life. Abnormal laryngeal voice quality in an infant may be a sign of airway obstruction, airway dysfunction, laryngeal manifestation of gastroesophageal reflux, or in some cases a more complex syndrome. Abnormalities of voice in an infant should trigger immediate referral for thorough evaluation. In later development, voice symptoms may also signal behavioral factors, such as voice abuse or compensatory strategies.

PREDICTING FUTURE SPEECH AND LANGUAGE PERFORMANCE

In this chapter, we have discussed the importance of recognizing early speech and language development in children with cleft palate. This is important for several reasons. First, babies are active learners, and a child with a cleft is developing speech and language in the presence of abnormal structure that is going to vary significantly among children. This variability will place demands that differ in type and magnitude from child to child and will therefore result in highly individualized communication strategies and communication impairments among children with clefts. Second, it is valuable to know a child's language age or stage of linguistic development for planning the timing of various forms of physical management, especially the timing of initial palatoplasty. Third, it is also valuable to be aware of early development in order to determine whether early speech and language intervention is required. Finally, with longitudinal information regarding early speech and language development important, information becomes available for attempting to predict later speech, language, and learning performance. In this era of diminishing resources, it is highly valuable to be able to identify which children we should be monitoring more intensively or which children should receive extra services with the goal of preventing some of the long-term impact on communication.

Therefore, a brief review of what is known about prediction of later speech and language characteristics in children with cleft palate is of value. Several predictors of later speech and language development have been identified in infants.[11,84] Chapman et al.[11] suggest that the presence of canonical babbling (i.e., consonant–vowel combinations) and the use of true consonants (i.e., consonants excluding /w/, /j/, and glottals) predicts better speech and language performance during the preschool period. These findings were replicated and extended in a study by Scherer et al.,[84] who found that a measure of babbling complexity known as Mean Babbling Level at 12 months predicted vocabulary size and speech accuracy at 30 months of age. These studies provide strong support for the need to consider early intervention to promote the foundation for later speech and language development. Additionally, the identification of different profiles of speech and language performance suggest that children with cleft palate, who also demonstrate receptive and expressive language delays, are at risk for later academic difficulty especially reading.[28,31] The presence of language impairment, even of a mild form, by entrance to kindergarten places the child at risk for academic delays. The association of language impairment with academic difficulties has such high predictive value that early monitoring and aggressive treatment of language impairment in the preschool period is recommended.

SPECIAL CONSIDERATIONS

As we have discussed, palatal clefting is often associated with other multiple malformation syndromes that may have communication impairment as one feature of the syndrome independent of the cleft palate and/or related to it. Cleft palate/craniofacial teams often serve as regional resource centers for complicated patients with known or suspected cleft palate and other physical impairments affecting the communication system, especially the speech production mechanism. It is not uncommon to have patients referred to specialists in cleft palate for consults regarding a variety of questions related to communication, especially to rule out cleft palate

or VPI. Frequently the cleft specialist is asked to assist in differential diagnosis or in treatment planning for these children with complex or challenging impairments that may or may not be associated with cleft palate. Therefore, in this chapter we would like to mention a few of these potentially challenging populations and some of the issues that the cleft provider may encounter.

''Is there a Submucous Cleft-Palate?"

One common and challenging patient is the child who has no or very limited speech production that is referred to rule out a submucous cleft palate or other peripheral, structural abnormality that might account for the lack of speech production. This is most often a very young child in the toddler/preschool age group or an early school age child who uses very limited recognizable, meaningful speech production, or whose speech production is completely unintelligible. These children may present with limited vocalization or extensive vocalization, but their sound repertoire is severely restricted. In many cases, their speech production is characterized primarily by vowels and the nasal consonants. This pattern often results in very "nasal sounding" speech, and hence the child is often referred to the cleft palate team to rule out submucous cleft palate or VPI. In many cases, there is significant hope among family and referring clinicians that the speech delay is due to a simple structural abnormality, such as submucous cleft palate, that can be corrected surgically and rapidly. However, thorough evaluation of the more global communication system in these children often reveals subtle or sometimes more obvious impairments in receptive and expressive language. In general, it is unlikely that an overt cleft palate, a submucous cleft palate, or even severe VPI in the absence of other factors would be the sole cause of such a communication disorder.

Once again, this discussion is more than academic. This is an area with great medical–legal ramifications that impacts not only the speech-language pathologist, but often the surgeons and other medical specialists involved with children with cleft palate as well. When these complex cases are referred to cleft providers, a thorough evaluation should be conducted including an in-depth history, cognitive-academic testing (including some neuropsychometric testing), detailed speech and language evaluation, and speech motor examination.[58]

Late Repair and Communication

Another population that has received little attention in discussions regarding speech and clefting is the recent immigrant or the child of a recent immigrant who presents to the cleft team with an unrepaired or partially repaired cleft. In many cases, these children are non-English speaking or speak English as a second language. These patients can be a special challenge for cleft teams but are especially challenging with respect to speech. One question that is frequently asked about older children or adults with unrepaired cleft palate is whether surgery will improve speech for these patients.

There are few studies on the speech outcomes for palatoplasty or secondary palatal management in patients who receive very late intervention. The few studies that are available are reports from work in developing countries where there has not been long-term follow-up or rigorous speech data. One exception is a report by Sell and Grunwell,[85] who evaluated the speech of 18 patients in Sri Lanka who underwent palate repair after the age of 11 years. The authors found that speech production was usually severely impaired in patients with such late repair, and the post operative results were variable and related to cleft type. Symptoms of hypernasality and nasal emission were improved with surgery, but there was little improvement in speech articulation when surgery alone was provided without speech therapy.

A study by Hall et al.[86] evaluated the outcome of secondary palatal management in adults. These authors suggested that symptoms of hypernasality can be successfully eliminated in adults with cleft palate. The authors suggest that patients should be thoroughly assessed prior to secondary palatal management for both velopharyngeal motion (especially lateral wall motion) and stimulability for articulatory improvement.

The most salient point that warrants mention in this chapter is that the speech of individuals who present at an advanced age with unrepaired palate or severe VPI requires in-depth investigation to determine the source of their speech symptoms. It is most likely that hypernasality and some patterns of nasal emission can be eliminated directly with surgical intervention. However, other articulation errors will require aggressive speech therapy. A thorough understanding of the profile of the individual's communication impairment will allow for realistic expectations from surgical intervention.

A related case that bears mention here is the child who is an active second language learner. There are anecdotal reports that suggest that many children who learn a second language after palate repair or after normalization of velopharyngeal function will demonstrate greater deficits in their primary language. There are no evidence-based studies, however, to document this relationship. One study in the literature describes two cases wherein the speech in two bilingual children was near normal in their second language following pharyngeal flap surgery while they continued to demonstrate numerous articulation errors including compensatory articulation errors in their primary language.[87] Therefore, it is important to consider both languages and phonetic/phonological systems when evaluating and creating treatment plans for bilingual speakers with cleft palate. It is also crucial that the families of bilingual children should be questioned carefully regarding the child's speech quality and intelligibility in their first language. It is sometimes true that the child's articulation skills in English (or the second language) are far superior to those in their first language. In some cases, the child may be experiencing severe difficulties especially in being understood within the family or first-language culture while appearing only minimally impaired in the second language in which clinical speech evaluations are being conducted.

Velocardiofacial Syndrome

Another complex but common patient population often referred to cleft specialists is the child with 22q11.2 deletion syndrome, or velocardiofacial syndrome (VCFS).[88] There is a full discussion of this syndrome in this text; however, communication impairment is one of the prominent features of VCFS and therefore warrants some discussion in this chapter.

Shprintzen et al.[88] first described the speech and resonance characteristics of children with VCFS. Goldin-Kushner et al.[89] identified significant language impairment in their population of children with VCFS. Gerdes et al.[90] reported on several measures of cognitive function in a group of preschoolers with VCFS and reported that 62% of the children tested were generally nonoral communicators at 24 months of age and these delays in language were beyond what would be expected for their developmental level. The early literature concerning the communication characteristics of children with VCFS has been largely composed primarily of descriptive, retrospective reports, clinical audits, case studies, and short summary statements.

Only one longitudinal study has been reported and there has been little information comparing the impairments observed in children with VCFS with other clinical populations. Scherer, D'Antonio, and Kalbfleisch[14] followed a group of children with VCFS from 6 months to 30 months of age and compared their communication development with that of comparison groups of children with cleft lip and palate, isolated cleft palate, and typically developing children. In this longitudinal study, the children with VCFS showed significant differences in receptive language, expressive language, and speech sound acquisition compared with the other three groups of children. This study suggested that the patterns of language and speech deficits in children with VCFS were not due solely to the effects of cleft palate or middle ear pathology associated with palatal clefting.

The speech patterns of children with VCFS have been described as having a predominance of glottal stop compensatory articulation substitutions. Reports suggest that 30–84% of children with VCFS have VPI.[91] Glottal stop substitutions appear particularly problematic because they occur for whole classes of sounds. Furthermore, it has been suggested that the high occurrence of glottal stop substitutions is responsible for much of the oral communication impairment in young children with VCFS. However, data from the longitudinal study reported by Scherer, D'Antonio and Kalbfleisch[14] did not support a simple relationship between the severe speech production abnormalities observed for the young children with VCFS and the presence of VPI. In fact, the VCFS group demonstrated significantly greater speech production deficits than children in the two cleft groups who also experienced VPI. The authors concluded that in young children with VCFS, the relationship between VP function and speech sound errors was not as simple and straightforward as has been suggested previously.

In a second study, D'Antonio et al.[92] compared the speech patterns of a group of children with VCFS to a group of children with speech impairment and some of the phenotypic characteristics of VCFS without a deletion at 22q11.2. The findings of this study showed that young children with VCFS had significant deficits in speech performance beyond that of the comparison group. Furthermore, the study demonstrated greater speech impairment in younger children with VCFS, such as smaller consonant inventories, greater number of developmental speech errors, greater severity of articulation disorder, and higher frequency of glottal stop substitutions, than in older children with VCFS or in the children without VCFS. As reported by Scherer et al., the relationship between ratings of velopharyngeal function and the speech variables analyzed in this study was not straightforward.

The data from these two studies has been interpreted to suggest that some children with VCFS demonstrate a communication profile that may be distinctive to this syndrome. To test this hypothesis Scherer, D'Antonio, and Rodgers[56] described the communication profiles of a group of children with VCFS compared with a group of children with Down syndrome. The profiles of the children with Down syndrome showed a flat profile, indicating all measures of communication were similarly delayed relative to chronological ages. In contrast, the children with VCFS showed vocabulary, pattern of sound types, and mean babbling length below cognitive and language age on other measures. The results suggested that communication profiles of children with VCFS differed qualitatively and quantitatively from children with Down syndrome and supported the hypothesis that some children with VCFS present with a profile of communication impairment that may be distinctive to the syndrome. The presence of this distinctive profile of communicative strengths and weaknesses points to the need for thorough assessment of cognitive and, language domains in addition to speech assessment in order to provide for adequate understanding of the disorder and for adequate intervention planning.

Finally, as discussed in the section on resonance and velopharyngeal function, there often is a pronounced need for diagnostic therapy as a means for providing information that will lead to accurate differential diagnosis for patients with VCFS. There are several sources of speech deviation in this population that can make differential diagnosis challenging. For example, it is a common clinical observation among speech clinicians experienced with the speech of children with VCFS that there is sometimes a disproportionate amount of perceived hypernasality in relation to velopharyngeal function as documented endoscopically, radiographically, and aerodynamically.[93] Especially in these cases of apparent disagreement among evaluation methods there is a greater need for stimulability testing and diagnostic therapy to assist in accurate diagnosis and treatment planning.

CONCLUSION

It is well known that the presence of a cleft palate may negatively impact an individual's ability to communicate effectively and may therefore cause significant social, emotional,

and educational hardship. Thus, the evaluation and management of communication disorders associated with cleft palate is a critical component of comprehensive cleft care.

Many discussions of communication disorders associated with cleft palate focus on articulation and velopharyngeal function. In this chapter, an attempt has been made to discuss the broader components of human communication and how they are impacted by clefting. Additionally, this chapter was written with the nonspeech pathologist in mind in an attempt to facilitate a more complete understanding and exchange between disciplines.

This chapter has called attention to the developmental course of communication and communication disorders associated with cleft palate. Additionally, we have stressed the importance of considering speech and velopharyngeal function as only one part of the broader communication process. Our discussion of the speech patterns associated with cleft palate emphasizes the view that speech is shaped by many dynamic linguistic processes that are active and that should be appreciated long before the first words are present in a child's communication process.

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