

**ASSOCIATED PROBLEMS IN CHILDREN WITH CLEFT LIP &
PALATE: A RETROSPECTIVE STUDY**

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RAVI KUMAR

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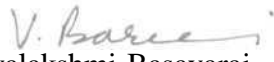
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APRIL 2007**

CERTIFICATE

This is to certify that this dissertation entitled "*Associated Problems in children with Cleft lip and Palate: A Retrospective Study*" is the bonafide work submitted in part fulfillment for the degree of Master of Science (Speech Language Pathology) of the student (**Registration No. 05SLP012**). This has been carried out under the guidance of a faculty of this institute and has not been submitted earlier to any other University for the award of any other Diploma or Degree.

Mysore

April, 2007


Dr. Vijayalakshmi Basavaraj
Director
All India Institute of Speech and Hearing
Naimisham Campus
Manasagangothri
Mysore-570 006.

CERTIFICATE

This is to certify that the dissertation entitled "*Associated Problems in children with Cleft lip and Palate: A Retrospective Study*" has been prepared under my supervision and guidance. It is also certified that this has not been submitted earlier in any other University for the award of any Diploma or Degree.

Place: Mysore
Date: April, 2007

M. Pushpavathi

Dr. M. Pushpavathi

Guide,
Reader,
Dept. of Speech Pathology
All India Institute of Speech and Hearing,
Mysore-570006.

DECLARATION

This is to certify that this dissertation entitled "*Associated Problems in children with Cleft lip and Palate: A Retrospective Study*" is the result of my own study under the guidance of Dr. M. Pushpavathi, Reader of Speech Pathology, Department of Speech Pathology, All India Institute of Speech and Hearing, Mysore, and has not been submitted in any other university for the award of any diploma or degree.

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*DEDICATED
TO THE MEMORY
OF
MY PARENTS*

*My Bhaiya
.....For What I am Today*

&

My Revered Guide

Dr. Pushpavathi, M.

*.....For her teaching, guidance, love and
affection*

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*"Gratitude is the hardest of all emotions to express.
There is no word capable of conveying all that one feels.
Until we reach a world where thoughts can be adequately express in words.
"Thank you (will have to do). "*

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language disorders. Other associated problems are hearing loss, dental problems, and mental retardation, with or without the presence of other syndromes.

Articulation disorders of varying degree of severity occur commonly in children with cleft palate. McWilliams (1953) & Spriestersbach (1956) described the articulation of heterogeneous groups of cleft palate individuals. Consonants misarticulated more than 60% times by children studied by Spriesterbach (1956) were /z/, /s/, /ʃ/, /ts/, /dz/, /s/. The consonants most correctly articulated at least 80% of the times were /m/, /n/, /j/, /ng/. The speech sound most frequently misarticulated by individuals with cleft palate is reported to be /s/ (Mc Williams, 1951, 1958).

Fletcher (1978) analyzed articulation data from 70 subjects with cleft palate. He classified the sounds into three categories- 1) sibilants: /s/, /z/, /ʃ/, /tʃ/, /dʒ/, (2) nonsibilant fricative: /θ/, /ð/, /v/, /f/, and (3) plosives: /p/, /b/, /t/, /d/, /k/, /g/. According to him, this classification resulted in more homogenous groupings of sounds than the more traditional assignment of sounds to fricative and affricative categories. He reported mean error percentages of 47 for sibilants, 24 for nonsibilants fricatives, and 17 for plosives, finding consistent with those of McWilliams (1953), Spriestersbach (1958) and Darley & Rouse (1956). His subjects misarticulated over 40% of the /s/ sounds studied in the initial, medial and final position of words. Spriestersbach et al., (1961) in a study of children between the ages of 3 to 17 years found more omission, distortion and fewer substitutions for blends than for single element.

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INTRODUCTION

Cleft is a failure of fusion of the medial nasal, lateral nasal and maxillary processes on one or both side. Cleft lip is a congenital deformity of upper lip, which varies from a notching to a complete division of the lip. It may be unilateral, bilateral or median cleft. Cleft palate is a congenital fissure in which it may extend through the uvula, soft palate and hard palate. It may be unilateral, bilateral or complete cleft palate. Cleft lip with or without cleft palate is the most common oro-facial anomaly in newborn infants, affecting one in every 750 live births.

The epidemiology and genetics of cleft lip and palate have been studied extensively in various countries by several investigators (Fraser, 1970; Derijcke et al., 1996). It is generally recognized that majority of these cases are consistent with multi factorial mode of inheritance (Fraser, 1970; Wyszynski & Beaty, 1996). Many teratogenic agents and factors in pregnancy are claimed to cause clefting (Wyszynsky et al., 1996) like cortisone (Yoneda & Pratt, 1982), anticonvulsants (King et al, 1996).

Cleft lip and palate are associated with several problems. The type of problem depends on the extent of the cleft and other associated factors like the timing of the surgery, presence of other birth defects etc. The associated problems may be divided into communication disorders and other problems. Communication disorders among cleft lip and palate individuals include speech disorders - articulatory disorder, voice disorder, resonatory disorders and

language disorders. Other associated problems are hearing loss, dental problems, and mental retardation, with or without the presence of other syndromes.

Articulation disorders of varying degree of severity occur commonly in children with cleft palate. McWilliams (1953) & Spriestersbach (1956) described the articulation of heterogeneous groups of cleft palate individuals. Consonants misarticulated more than 60% times by children studied by Spriesterbach (1956) were *IzI*, */S/*, *IQI*, */ts/*, */dz/*, */s/*. The consonants most correctly articulated at least 80% of the times were */ml*, *In/*, */j/*, */ng/*. The speech sound most frequently misarticulated by individuals with cleft palate is reported to be *I si* (Mc Williams, 1951, 1958).

Fletcher (1978) analyzed articulation data from 70 subjects with cleft palate. He classified the sounds into three categories- 1) sibilants: *Is/*, *IzI*, *HI*, *I tf/*, */(£/*, (2) nonsibilant fricative: *IQI*, *Id/*, *hi*, *If/*, and (3) plosives: *Ipl*, *Pol*, *IM*, *Id/*, *Ikl*, *Igl*. According to him, this classification resulted in more homogenous groupings of sounds than the more traditional assignment of sounds to fricative and affricative categories. He reported mean error percentages of 47 for sibilants, 24 for nonsibilants fricatives, and 17 for plosives, finding consistent with those of McWilliams (1953), Spriestersbach (1958) and Darley & Rouse (1956). His subjects misarticulated over 40% of the *I si* sounds studied in the initial, medial and final position of words. Spriestersbach et al., (1961) in a study of children between the ages of 3 to 17 years found more omission, distortion and fewer substitutions for blends than for single element.

Cleft palate in association with velopharyngeal incompetency is more likely to interfere with some manner of articulation than of others. Van Denmark (1979) observed a higher rate of misarticulation of fricatives and affricatives than of plosives in subject with velopharyngeal dysfunction. Pitzner & Morris (1966) found that cleft palate individuals with poor velopharyngeal closure were more likely to misarticulate /r/ and /l/ sound than of cleft palate subjects with good velopharyngeal function.

Moll (1968) reported that velopharyngeal incompetence is more likely to interfere with manner of articulation than with others. He concluded that tongue tip complex sounds include fricatives and affricatives which involve lingual contact tended to be more defective than those involving only the lips. Individuals with cleft also appear to misarticulate voiceless sound most frequently than the voice cognates (Spriestersbach et al., 1956; McWilliams, 1953, 1958). Sherman et al., (1959) found that glottal stops were used more frequently to replace voiceless than voiced consonants. Spriestersbach et al., (1961) found that voiced stops and affricatives were better articulated than their voiceless counterparts.

Voice disorders in subject with cleft palate may be classified as phonatory disorder and resonatory disorder. Children with cleft palate reported to have unintelligible speech due to hypernasality. Velopharyngeal dysfunction is the main cause leads to the hypernasality. Individuals with cleft palate and velopharyngeal incompetency have traditionally been described in the literature as having a hypernasal voice quality. Hairfield et al., (1988) reported that 68% of 85 children and adults with cleft lip and palates were oral or mixed oral-nasal breathers, whereas only 32% were nasal or predominantly nasal breathers. The

degree to which hypernasality is perceived by a listener will depend on the characteristics of the entire vocal tract and not only on the size of the functional opening in the velopharyngeal valve (Curtis, 1968).

McDonald & Baker (1951), Westlake & Hess (1959) recognized faulty phonation as an important attribute of the speech of cleft palate population. Bzoch (1964) reported hoarseness and breathiness in cleft palate patient. Reduction in loudness is often a characteristic of people with velopharyngeal dysfunction. (McWilliam & Phillips, 1979). Counihan & Cullinan (1972) found lowering of nasality rating with increasing vocal intensity in cleft palate and normal males. McDonald & Baker (1951) mentioned hoarse voice as abnormal quality feature in cleft palate voice.

Limited attention has been paid to the language aspects compared to the articulatory proficiency and voice in the cleft palate subjects. Bzoch (1966) indicated that more than 50% of children, he studied, had delayed language development. Spriestersbach et al., (1958) reported that shorter mean length of utterances (MLU) in cleft palate children than normal children of the same age and gender.

Nation (1970) used the Peabody Picture Vocabulary Test (PPVT) to assess both comprehension and usage on 25 children with cleft and concluded that the presence of cleft affects development of vocabulary. Smith & Mc Williams (1968) administered ITPA to 136 children with cleft lip and palate and reported that particular weakness in vocal expression, gestural output and visual memory. Bzoch et al., (1973) studied twenty-five infants with cleft lip and palate and

concluded that consistent and significant delay in expressive language skills. Pannbacker (1975) reported that cleft subject were inferior to normal on mean length of utterances.

Other Associated Problems

Individuals with a history of cleft palate are at high risk for otitis media and associated conductive hearing loss. This is due to malfunction of Eustachian tube (Bluestone & Doyle, 1988), which connects the middle ear, to the posterior pharynx.

Trisomy 13 is a lethal chromosomal disorder. The incidence of this disorder is about 1 in 5000 live births (Jones, 1997). Many infants with trisomy - 13 have a midline cleft lip and palate facial anomalies

Children with cleft palate are significantly impaired in their cognitive ability compared to normal children. The impairment is more in the area of verbal intellectual skills (Goodstein, 1961).

Dental problems in children with cleft palate are reported to be more compared to normal children. The dental problems include missing teeth, supernumery teeth, open bite and under bite etc (Proffit & White 1991)

Need for the study

There are many studies done in the western context to analyze the associated problems in unrepaired cleft palate population. But there is no

REVIEW OF LITERATURE

"Nature, as we often say, makes nothing in vain, and man is the only animal who he has endowed with the gift of speech. And whereas mere voice is but an indication of pleasure or pain, and is therefore found in other animals (for their nature attains to the perception of pleasure and no further), the power of speech is intended to set forth the expedient and inpedient, and therefore likewise the just and unjust.

Aristotle, Politics, BOOK I (7).

Speech is unique to human being and it is a complex human behavior. It can be defined as a genetically determined individual psycho-physiological activity consisting of the production of phonated, articulated sound through the interaction and coordination of cortical, laryngeal and oral structure (Newman, 1963). Although, it can be developed spontaneously in human beings, development of speech depends on many factors like adequate function of CNS, adequate feedback mechanism, adequate intelligence, adequate sensory motor development, structure and function of the oral cavity. Defect at any of these levels lead to the speech disorder. Defect in oral cavity is one of the condition which leads to abnormal speech. Cleft lip and palate are congenital malformations that occur in utero and are associated with many problems.

The term "Cleft" is defined as an opening in an anatomical part that is normally not open. A cleft is a lack of union of embryonic oral and facial elements. There has been some argument as to whether this lack of fusion is caused by a failure of embryonic parts to grow towards each other (Fraser, 1961)

or a ruptured of already fused elements (Kitamura, 1991). Cleft is a failure of fusion of the median nasal, lateral nasal and maxillary processes on one or both side. A cleft condition is determined during the fourth to eighth week of pregnancy. After this critical period, nothing the mother does can cause a cleft and nothing a mother does can avoid the cleft. Sometimes it is determined even before the mother is aware that she is pregnant.

The lip is normally a solid structure spanning the entire distance from one corner of the mouth to the other corner. A cleft lip (which almost always refers to the upper lip; cleft of the lower lip are extremely rare) is an opening in the lip so that the lip is not contiguous. If the cleft is incomplete, it may be only a minor notch or may extend almost to the nostril. If it is complete, it will include entire lip and continue into the floor of the nostril. A cleft lip may be unilateral (commonly occur on left), only one side is affected, or bilateral, both sides are affected. Similarly, a cleft of the palate, which is normally intact, is an opening in the palate such that there is a continuous passage between the mouth and the nose because the palate serves as both the roof of the mouth and the floor of the nose.

The anterior two-thirds of the palate make up the hard palate, commonly referred to as the roof of the mouth. The bone under the mucosa is composed of maxillary and the palatine bones of the skull. The soft palate or velum consists of the posterior one-third of the palate. This structure has no bony underlay and is composed of muscles and mucosa. A complete cleft of the lip and palate extends from external lip and palate extends from the external lip posteriorly through the alveolar arch and the hard and soft palate. The soft palate and the uvula are split. The nasal septum is usually attached to the larger of the two palatal segments in

the unilateral clefts but is not attached to either segment in bilateral cleft. Isolated cleft palate, without cleft lip, and also varies in severity. It may include the entire hard palate posterior to the incisive foramen and the soft palate; it may involve only a small portion of the posterior part of the soft palate; or it may be between these two extremes.

In submucous cleft palate, the palate appears to be structurally intact, but there usually are both bony and muscular deficits (Weatherley-White et al., 1972; Velasco et al., 1988). The defect is associated with a triad of symptoms including a bony notch in the posterior border of the hard palate, a bluish line at the midline of the soft palate, and a bifid uvula. The notch or bony cleft is not always observable during routine examination but can often be detected by manual palpation. The submucous cleft of the hard palate is not functionally significant, but the muscular deficit found in the soft palate. The muscular cleft, covered only by oral mucosa, is seen as a bluish line through the length of the soft palate. The incidence of submucous ranges from 5-10% of cleft palate population. (Weatherley-White et al., 1972).

Congenital palatopharyngeal incompetence (CPI) was described by Calnan (1971a) & Croft et al. (1978). This condition is present at birth which results in velopharyngeal dysfunction for speech. Velopharyngeal insufficiency means that the velum and pharyngeal muscles donot or can't, for some reason, produce the optimal closure between oro- and naso- pharynx. This defect is usually diagnosed after speech develops (Morris et al., 1982) and hypernasality is heard. A number of anatomical and physiological conditions lead to velopharyngeal dysfunction. They include congenitally short palate, reduced

palatal bulk, deep or enlarged pharynx, malinsertion of levator muscles and combination of these. VPD is a major contributory factor to the speech and language problem associated with cleft palate, other factor such as faulty learning parental attitudes and reactions, otological problem, and negative self- evaluation must be considered (Richman and Eliason, 1986).

Both cleft lip and cleft palate alone are etiologically heterogenous. There is neither a single cause nor any single etiological model that explains the occurrence of either type of cleft. It is generally recognized that a majority of these cases are consistent with the multifactorial modes of inheritance (Fraser, 1970; Demenais et al., 1984; Wyszynski & Beaty, 1996). Several reports have shown that supplementation of some vitamins like B6 and folic acid during the first trimester in women who had previously borne offspring with cleft lip and palate has a protective effect, with a marked decrease in recurrence rate (Cohlan, 1953; Peer et al., 1958; Yoneda & Pratt, 1982). Many teratogenic agents and factors in pregnancy are claimed to cause clefting (Wyszynky et al., 1996). cortisone (Diewert & Pratt, 1981), anticonvulsants (King et al, 1996), salicylates (Saxen, 1975a, b), vitamin A (Cohlan, 1953), aminopterin (Fara, 1968), organic solvents (Laumon et al., 1996), maternal smoking (Kallen, 1997), maternal hypoxia (Millicovsky & Johnston, 1981), maternal rubella (Peer et al., 1958), maternal diabetes mellitus (Peer et al., 1958) and season of gestation (Fraser & Calnan, 1961; Rintal & Stegars, 1982) are some of these agents and factors. A small proportion of clefts may be part of genetic syndromes with Mendelian inheritance of other recognized syndromes, or of unclassified multiple malformations (Melnick et al., 1980a, b; Demenais et al., 1984; Stoll et al., 1991).

It is also suggested that cleft lip with or without cleft palate and isolated cleft palate are separate genetic disorders (Fraser, 1970). Some of the syndromes in which a cleft is a feature are known to be caused by an abnormal gene. Examples of single-gene disorders that include clefts are Treacher Collins syndrome, velocardiofacial syndrome, Stickler syndrome, and Van der Woude syndrome, all of which are autosomal dominant.

The following review section highlights the studies done in the area of epidemiology and associated problems in children with cleft lip and palate.

I. Epidemiology

Many epidemiologic studies have been conducted on the incidence of cleft lip, cleft palate, and cleft lip and palate in the United States and other countries. The results show a wide variation in the risk of developing clefts within and among races. A safe conservative estimate of incidence appears to be approximately one in every 750 live births, although there are some indications that the rate may be even higher. Rintala (1983) reports an incidence rate of 1/462 in Finland & Jensen et al (1988) reports 1/529 in Denmark. Hook (1988) reviews two sets of data from Hungary (Czeizel, 1984) and the United States (Myriantopoulos & Chung, 1974) reports overall rates of one in 645 and one in 625 respectively. It has been repeatedly shown that individuals with cleft of the palate only are far more likely to have associated anomalies than are individuals with clefts of the lip (Shprintzen et al., 1985b; Womersley & Stone, 1987).

Subjects with cleft palate also have a higher percentage of siblings with malformations than do patients with cleft lip (Meskin & Pruzansky, 1969).

Females have a higher rate of associated anomalies than do males, regardless of the type of cleft (Meskin & Pruzansky, 1969). Two studies (Myriantopoulos & Chung, 1974; Siegel, 1979) reported a higher incidence of associated anomalies in blacks than in Caucasians. In their study of 1000 patients with cleft Shprintzen et al. (1985b) reported that one or more associated anomalies in 44.6% cleft lip only, 50.3% of those with cleft lip and palate, 67.9% of those with cleft of the secondary palate only, and 76.8% of those with submucous cleft for an overall prevalence of 62.4%. Iregbulem (1982) reported that there were eight cases of clefts of the lip or palate or both giving an incidence of one in 2,703 live births in Nigeria and he found that the frequency of cleft lip only was 49%, of cleft palate only was 19%, and of cleft lip and palate was 32% and the proportion of adults with unoperated clefts was high. Associated congenital abnormalities observed in 18% of the patients.

Gregg & Boyd et al, (1994) reported that the incidence of the various types of cleft lip and/or palate drawn from a regional database of all affected children born in Northern Ireland during the period 1980-1990. The incidence of these anomalies was 1.28 per 1000 live births (1:781). 53% of clefts involved the secondary palate only, 16 % the primary palate only, 26 % involved both primary and secondary palate and 5 % were unconnected. Overall, more males than females were affected and there were more males than females in the group having complete clefts. Separate clefts of the lip and palate occurred exclusively in males with only one exception. Unilateral clefts were more common on the left side. Within the group showing complete unilateral cleft of the primary and secondary palate, left- sided clefts were more commonly male, right — sided clefts

were more commonly female. There were no statistically significant sex differences between sides in the unilateral primary palate cleft group.

Shapira, Lubit, et al., (2000) studied on the frequency and patterns of distribution of cleft lip, cleft lip and alveolus, cleft lip and palate and isolated cleft palate, together with possible association between sex, type of cleft and affected side more studied from records of 278 individuals with clefts from four cleft centre in New York city area. The type of the cleft varied between sexes. Males reported to have significantly higher rates of cleft lip and palate ($p < 0.0001$) and females had higher rates of isolated cleft palate ($p < 0.0001$). No sex differences were found for cleft lip or cleft lip and alveolus. Unilateral clefts of both the primary and secondary palates were found to occur over three times more frequently than bilateral clefts and left side predominance was demonstrated.

Machabova, Bansky, Guzanin, et al., (2006) studied a retrospective active survey collecting clinical data of children with orofacial clefts examined and operated on in the three main specialized departments of plastic surgery in the Slovak Republic over 16 years (1985-2000) revealed total incidence of 1.61/10(3) live births (LB). 1,849 children suffering from orofacial clefts were recognized out of 1,147,236 live births. Total incidence of orofacial cleft per 1,000 live births was determined by types, gender, regions, districts and seasonal variation. The highest rate, 40.5% of clefts, affected the primary and secondary palate, more than 32% were of cleft palate type and about 26% cleft lip and about 1% of associated malformations. Clefts in males were significantly more common than in females.

II. Associated Communication Disorders

When a child is born with a craniofacial deformity that involves the oral structures which may or may not be accompanied by other factors of developmental delay, the acquisition of speech and language becomes a much more difficult process. Babies born with abnormal speech mechanisms are at high risk for the development of disordered speech (O'Gara & Logemann, 1988). The primary role of speech language pathologist is to assist the child with a craniofacial anomaly to achieve his/her maximum communication potential. It is widely accepted that communication skills begin to emerge in the first few weeks of life (Bloom & Lahey, 1978; Dore, 1986).

A few investigations concerning the speech characteristic associated with extent of the isolated cleft palate have been reported (Jacobsson et al., 1990; Marrinan et al., 1998). In a study of Jacobsson et al., (1990), patients were divided into three subgroups depending on the extent of the cleft (complete, partial, soft only), while Lohmander-Agerskov et al., (1993) and Marrinan et al., (1998) had two subgroups (hard and soft palate, soft palate only). Jacobsson et al., (1990) found that patients with a cleft in the soft palate only showed significantly less articulation errors than patient with clefts that extended into the hard palate. In addition, Lohmander-Agerskov et al., (1993) found a significantly higher amount of compensatory articulation, in this case defined as velar or palatal articulation of dental consonants, among the group with a cleft in the hard and soft palate. A relationship between cleft type and language and learning disability has been suggested since language delay (Richman et al., 1986; Eliason, 1990) and a higher

rate of reading disability (Richman et al., 1988) were observed among children with a cleft palate only than among children with a cleft lip and palate.

Communication problems resulting from a variety of causes and combination of causes occur in most, but not all, patient with craniofacial abnormalities. The communicative disorder frequently evidenced by patient with craniofacial anomalies include resonance disorder such as hypernasality, hyponasality, and cul de sac resonance; speech sound production disorders including compensatory articulation errors such as glottal stops and pharyngeal fricatives , nasal emission, weak consonant production, omissions, substitutions, and distortions; voice disorder such as hoarseness and weak intensity; and language and learning problems (Peterson-Falzone, 1986; Trsot-Cardamone, 1990).

Cohen & Bankier (1991) estimated that there were more than 340 known syndromes involving oro-facial clefting. Although velopharyngeal inadequacy (VP1) is undoubtedly a major contributory factor to the speech and language problems associated with cleft palate, other factor such as faulty learning, parental attitudes and reactions, otological problems, and negative self-evaluation must be considered (Richman & Eliason, 1986).

Language disorders in children with craniofacial syndromes occur with greater frequency than in a normal population. Hearing impairment is found frequently in both children and adults (Selder, 1973). Crisdale (1981) pointed out that congenital conductive hearing loss is an intrinsic component of many craniofacial syndromes. Some of these losses are related to microtia, whereas

others represent what he calls "invisible" deformities that results in a delay in identification. Some of these hidden anomalies are ossicular deformities, stapedial fixation, or tympanic membranes that are small and oblique with abnormal landmarks.

Mental development is often a major issue in the diagnosis and the outcome of major craniofacial abnormalities. Nearly half of the syndromes listed by Siegel-Sadewitz & Shprintzen (1982) are reported to be frequently or probably associated with cognitive disorders. Some syndromes are associated with malformations of the brain resulting in mental retardation in 100% of affected children. So, Communication disorders among cleft lip and palate individuals include (a) Articulation disorder (b) Voice disorder (c) Resonatory disorder and (d) Language disorders and other associated problems which are hearing loss, dental problems, psycho-social factors and mental retardation and with or without the presence of other syndromes.

II. a. Speech Disorders

(i) Articulation Disorder: Misarticulation is an important speech disorder in many persons with cleft palate. Formation of the sounds of speech is easily influenced by deviations in oral structures and functions (Witzel et al., 1980; Vallino & Thomson, 1993). So that the risk of articulation problem is high in those with cleft lip and /or palate or other craniofacial abnormalities. Speech sounds errors are classified as phonetic or phonologic errors by the speech language pathologist. Phonetic errors in the articulations or formations of sounds of speech are related to abnormalities in anatomy, function, and/or motor control

in learning. Phonological errors indicate a difficulty in the child's organization, learning and representation of the sound units and sound system. As a group, these children achieve articulation test scores below available norms or demonstrate poorer articulation than age- matched controls (Bzoch, 1965; Van Denmark, Morris & Vandehaar, 1979). Even children and adolescents with adequate velopharyngeal function often demonstrate poorer articulation skills than their non-cleft peers (Fletcher, 1978).

Speakers with cleft palate have demonstrated that omissions and substitutions occur frequently than other types in young children. (Bzoch, 1956, Spriestersbach et al., 1956), although errors related to distortions occur most frequently in older children and adults (Bardach et al., 1984; Van Demark, 1966). The omissions observed in young children with cleft palate often reflect normal developmental processes (i.e., final consonant deletion, weak syllable deletion) that might be expected in any child. Although nasal distortion is probably one of the most salient characteristics of speech associated with cleft palate, research findings indicate that misarticulation related to oral distortion occur far more frequently than those attributed to nasal distortion (Bzoch, 1956; Van Demark, 1964). Bzoch (1956) observed that nasal distortion persists with age, whereas other distortions tend to decrease in frequency of occurrence.

Numerous investigations have demonstrated that children with cleft palate have more difficulty producing pressure consonants than other classes of consonants (McWilliams, 1953; Van Demark, 1969; Morris & Vandehaar, 1979). They typically misarticulate fricatives and affricates most frequently, followed by plosives, glides, and then nasals. Velopharyngeal inadequacy is generally

considered to be a major factor responsible for errors on the former three classes of sounds because each requires high intraoral air pressure. Consonants involving lingual contacts were found to be more defective than those involving only the lips across all manner of production categories.

In two descriptive studies, McWilliams (1953) and Spriestersbach et al., (1956) described the articulation of heterogenous groups of adults and children with palatal clefts. Consonants misarticulated more than 60 % of time by the children studied by Spriestersbach (1956) were *Iz*, *Isl*, /tʃ/, /ts/, /θ/, /tʃ/, /dʒ/, /ð/, /s/, and /tʃ/. The consonants correctly articulated at least 80% of the times were /m/, /h/, /n/, /j/, and /r\l/. In the McWilliams study, the most frequently misarticulated sounds were *Isl* (63%), /z/ (61%), /d/ (48%), and /tʃ/ (44%). Only 11% of the /p/ sounds sampled and 9% of the *Pol* sounds were misarticulated. The *Isl* is the speech sound most frequently and most consistently misarticulated by individuals with cleft palate (McWilliams, 1953, 1958). Fletcher (1978) reported that his subject misarticulated 46%, 43% and 49% of the /s/ sounds studied in the initial, final and medial positions of the words respectively.

Individuals with cleft appear to misarticulate voice sounds more frequently than the voiced cognates (Spriestersbach et al., 1956; McWilliams, 1958). They studied on articulation of a group of children with cleft lip and palate. They took the speech sample of 25 children (9 girls and 16 boys) in the age range from 3 years 7 months to 8 years 3 months born with cleft lip and/or cleft palate and they analyzed in order to present the following: - i) a systematic description of their articulation skills, ii) An estimate of possible retardation of their ability, and iii) a

description of their patterns of their patterns of misarticulation. The sample was restricted to children between the ages of three and eight who singletons were and of the Caucasians race with normal hearing. Templin articulation test was administered. The subjects used their study had little difficulty in the articulation of vowels and diphthongs. The mean percentage of correct production of the vowels and diphthongs for the group as a whole was 96%. The consonants misarticulated more than 60% of the time by this group in order of decreasing defectiveness are /z/ /θ/, /s/, /tʃ/, /dʒ/, /ð/, /ʃ/, and /t/. The five consonants giving this group the least difficulty in order of increasing defectiveness are /m/, /n/, /h/, /l/, and /r/. These children also had significantly less difficulty with voiced consonants than they did with voiceless consonants. As a group, the cleft palate children tested had least difficulty with the nasal consonants and the greatest difficulty with the fricative consonants. Omission type errors were observed most frequently followed by substitution and distortion in that order. The subjects had significantly less difficulty producing the consonants correctly when they appear as singles than when they appeared as elements of blends.

Sherman, Spriestersbach & Noll (1959) studied glottal stops in the speech of children with cleft palate. They recorded speech sample each 30 second long, consisted of nursery rhymes, counting story telling, reading and conversation from the speech of 50 cleft palate children and 50 children with functional misarticulations in the age range of four to ten years. The cleft palate samples were rated more severely, on the average, than the functional misarticulation samples in both cases. According to frequency counts based upon phonetic transcription, glottal stops occurred more frequently, on the average, in the cleft

palate samples. For both groups of samples, most of the glottal stops occurred as pre-vocalic intrusions or substitutions for voiceless consonants, the latter being almost entirely stop plosives. Although glottal stops were more conspicuous and more frequent in cleft palate samples than in the functional misarticulation samples, there was considerable overlap of the two distributions for both the frequency and the severity measures. They found that glottal stops were used more frequently to replace voiceless than voiced consonants.

Van Demark & Van Demark (1967) compared the articulation of children with cleft palate who had good velopharyngeal function and children with functional articulation disorder. A standard sample of connected speech was obtained from 28 children with articulation disorder considered to be functional and 33 children with a physically managed cleft palate who could achieve a manometer ratio of 1.00. The speech samples were analyzed for articulation errors and a group of judges rated the samples on the basis of articulation defectiveness and severity of nasal voice quality. The result of the articulation analysis showed that the functional articulate group made significantly more glide sounds than did the cleft palate group. There were no other differences between the two groups in the total number of errors, the number of errors in manner of production or in type of misarticulation. There were significant differences in judges rating of articulation defectiveness and nasal voice quality. It appears that when a child with cleft palate achieves velopharyngeal closure, it is difficult to differentiate his speech from that of a child with a functional articulation disorder.

Philips & Harrison (1969) conducted a cross-sectional study intended to describe speech maturation in 74 children between the ages of 24 and 72 months

with the cleft palate only, but all cleft involved the soft palate and to compare their maturation with that of 127 normal children between the ages of 30 and 72 months who were attending nursery and kindergarten programs located in metropolitan community. They reported that the children with clefts made many more articulation errors than did normal children. They noted that the articulation scores of 94 % of pre-school children with cleft palate aged 3 years and older were inferior to the mean articulation scores of the normal children of the same ages. This finding supports a rationale for providing a program of speech stimulation for all cleft palate infants when the palatal defects involve the soft palate.

Bless et al., (1978) commented that individuals with velopharyngeal incompetence are most likely to misarticulate consonants that are adjacent to nasal sounds, whereas individuals with marginal velopharyngeal incompetence may nasalize oral consonants adjacent to nasals through assimilatory relationships

Logemann (1983) stated that children with cleft palate who are between 24 months and 5 years old tend to have articulatory placement errors but correct manner of articulation. They maintain manner by placing articulation posterior to fistulas or velopharyngeal opening. Older children present accurate articulatory placement but manner errors in the form of faulty release of stops, fricatives and affricates.

Peterson-Falzone (1990) conducted a cross-sectional analysis of speech for 240 children (age 4 years to 10 years 11 months) with repaired cleft palate. More than 90 % of the young school aged children she studied demonstrated articulation problems related to place or manner of production. Approximately 17 % exhibited

consistent audible nasal emission with associative hypernasality. According to Peterson-Falzone (1990) only 3% of the children she studied demonstrated speech that was entirely asymptomatic. It is noted that those patients who had received secondary surgical/prosthetic management for VPI were excluded from this study during subject selection. It is also noted that many of the patients' studied by Peterson- Falzone had received primary palatal surgery. Outside the United States and had not received routine team management. This findings account for the high prevalence of articulation problems observed in her school aged group.

Dalston (1990) described the communication skills of 63 children (age 3-5.11 years) and 36 adolescents (age 14 - 15 years) with repaired cleft palate. Seventy five percent of the younger patient in his study and 25% of the older patient demonstrated some type of communicative disorder. Articulation disorder was noted in 74 % of the 4- to 5-year-old children and in 14 % of the adolescents

Lohmander-Agerskov, Frieda & Lilja, et al., (1994) analyzed for place and manner of articulation of pre-speech in 35 children with clefts and palate or palate only. Transcriptions were made from tape recorded babbling sequences. The frequency of selected phonetic features was calculated. The results showed that anteriorly placed sounds (i.e., bilabial, dental and alveolar sounds) occur frequently among the children with cleft palate only and in the non-cleft children. In children with cleft lip and palate, posteriorly placed articulations predominated.

(ii) Voice Disorder/ Phonatory Disorder

Voice production is a highly integrated, automatic behavior that requires the finely tuned interaction of the respiratory system, the laryngeal structures, and

the supraglottic vocal tube with both its oral and nasal branches. "Phonatory disorders" refer to a problem that occurs at the level of larynx. Hypernasality and other disturbances occur supraglottally which referred to as a "resonance disorder."

McDonald & Baker (1951), Westlake & Hess (1959) all recognized faulty phonation as an important attribute of the speech of the people with palatal clefts. McDonald & Baker (1951) pointed out that laryngoscopic examination often reveal "hyperemia" and "hyperplasia" of the vocal folds in adult patients. The occurrence of phonatory disorder in individual with clefts is not well understood because it may vary from one sample to another, depending on such variables as velopharyngeal adequacy and the criteria for determining the presence of phonatory deviation. It is well clear that phonatory disorders are more common in this population than in non- cleft individuals.

Brooks & Shelton (1963) reported an occurrence of 10% of 76 children while Takagi et al. (1965), in a retrospective study, found only 0.6% in 1061 cleft patients over a wide age range. Marks et al. (1971) identified laryngeal dysfunction in 34% of 102 subjects over 6 and under 20 years of age. More males than females were thought to have phonatory deviations. Deering (1984) investigated the prevalence of dysphonia in 38 subjects with clefts and found that 50% had aberrant phonation. Of these, 23.7% were described as hoarse, 13.2% as harsh and 10.5% as breathy.

In 1969, Mc Williams et al. provided information about 43 children with cleft palate. Thirty-two of the 43 children with chronic hoarseness were

successfully laryngoscoped, and 84% had positive vocal-cord findings. The most usual pathological condition was bilateral vocal-cord nodules, which occurred in 23 or 71.9% of the children. Other pathological conditions included posterior glottal chink, bilateral vocal cord hypertrophy, slight anterior edema, and improper approximation of the vocal cords. Because of loss of pressure through the velopharyngeal port, some cleft palate patients have difficulty creating voice of sufficient loudness for conversational purposes

Bzoch (1979) said that "dysphonic characterized by aspirate voice." He defined the dysphonia as a habituated laryngeal voice abnormality caused by the adjustment of intrinsic muscles of larynx to produce a partial opening of the glottis during voice production. It is considered as a laryngeal compensatory voice disorder separate from hoarseness or roughness of the fundamental frequency and may be confirmed as a functional voice disorder only when otolaryngological examination indicates clearly that there is no muscular weakness or lesion in the vocal folds or other structural abnormality limiting the abduction of the vocal folds. The use of weak and aspirate voice was identified as categorical aspects of abnormal speech behavior in 313 of the 1,000 cleft palate studies. Trained judges can agree on classifying a voice as aspirate by simply listening to samples of the patient's conversational speech. By paying careful attention to the breathing pattern during conversational speech and by timing the number of seconds a subject can sustain phonation of isolated vowels, aspirate phonation can be identified. Subject with habituated aspirate voice were most often unable to sustain vowels for 10 seconds as more on three trials for each vowels. Aspirate phonatory voice quality appears often to be developed by these children early in

order to improve the intelligibility of their speech when they must communicate while they have velopharyngeal insufficiency. The distortion from velopharyngeal incompetency due to both nasal emission and hypernasal resonance characteristics is greatly increased perceptually when patients speak with clear phonation as contrasted to weak aspirate phonation. Strangled voice appears to be associated with an attempt to be non-nasal in the presence of velopharyngeal incompetence of sufficient magnitude to lead inevitably to hypernasality.

D'Antonia, Marsh, Province, et al., (1988) studied on the prevalence of laryngeal/ voice findings in a group of 85 patients referred for multimethod evaluation of velopharyngeal dysfunction. Forty-two patients had overt clefts of the hard palate, soft palate or both 10 patients had submucous cleft palate and 33 patients had no evidence of clefting. Fifty patients were male and 35 were female and their age-range was from 3 years to 52 years. Forty-one percent of patients had auditorily perceived voiced symptoms and/or observable laryngeal abnormalities. Twenty-one percent of patients had vocal fold nodules or thickened vocal folds. There was no clear relationship between laryngeal/ voice findings and estimated subglottal pressure. Patients with laryngeal/ voice findings (with or without nodules) had average estimated subglottal pressure values which were outside the normal range more often than patients without laryngeal/ voice findings. These results suggest patients referred for assessment of velopharyngeal dysfunction should receive a comprehensive evaluation which includes screening laryngeal structure and function.

Bressmann, Sander & Horch et al., (1998) conducted a screening study for voice problem in cleft lip and palate (CLP) patients in German speaking area. 154

patients with cleft lip and palate were examined. Voice quality was assessed perceptually according to the rbh- system (i.e. roughness, breathiness and hoarseness). Supra segmental and nasal resonances were also assessed. Using a new computer programmed for the apparatusive analysis of voice quality, data for pitch, intensity and perturbation (jitter and shimmer) were obtained for 4 sustained vowels. Based on the perceptual analysis, they found that the prevalence of severe voice disorders in cleft palate patients was 6.5% which is lower than reported in most other studies. The incidence of voice disorder is slightly higher than in normal population. Since the prevalence of severe voice disorder was low, none of the perturbation quotients differentiated between modal and disordered voices. A weak correlation was found between jitter and the rating for hoarseness.

Hocevar-Boltezar, Jarc & Kozelj (2006) investigated to compare the prevalence of ear, nose and particularly voice problems in groups of children with cleft palate and unilateral lip, alveolus and palate. On the basis of history, regular otorhinolaryngological examinations and hearing tests, the prevalence of different pathologies was assessed in 80 cleft palate children (35 boys and 45 girls) and 73 unilateral cleft lip and palate children (47 boys and 26 girls). Ear pathology was reported in 53.8 % of cleft palate children and in 58.9% of unilateral cleft lip and palate children. Nasal breathing was reported impaired in 17.5 % and 49.3% unilateral cleft lip and palate children. Dysphonia was detected in 12.5 % of cleft palate and 12.3 % of unilateral cleft lip and palate children. In 9.2% of all cleft children, functional voice disorder caused a hoarse voice.

(iii) Resonance disorder

Resonance, an acoustic phenomenon, is a complex attribute of speech that is not completely understood. It may be defined as "the vibratory response of a body or air-filled cavity to a frequency imposed upon it." (Wood, 1971). The term "hypemasality," "nasalization" and "nasalance" also appear with reference to disorders marked by an increase in nasal resonance, which is a major characteristic of speakers with velopharyngeal incompetence. Speech is not outstanding in any of its resonance characteristic depends upon the integrity of the vocal tract, including the supraglottal structures. For example, too much nasal resonance coupled with reduced oral resonance increases the perception of nasality, whereas an increase on the oral side may serve to decrease it. Hypemasality occurs when the oral and nasal cavities are coupled when they should not be. The result is that the sound wave is diverted into the nasal airways, and speech sounds as if it is coming through the nose. The primary cause of hypemasality is the coupling of the oral and nasal cavities regardless of how the phenomenon may be displayed instrumentally.

Hypemasality and nasal emission are both speech characteristics associated with poor velopharyngeal structure and function. Nasal emission of air may be associated with reduced oral breath pressure for pressure consonants and the combination of hypemasality and reduced oral breath pressure may mask place and manner of articulation faults (Wells, 1971). Speakers who do not achieve sufficient velopharyngeal closure for production of the pressure sounds may demonstrate visible nasal escape from one or both nostrils for obstruents.

This escape, although inaudible, may fog a cold mirror held at the nose and thus is sometimes referred to as "visible" nasal emission.

Audible nasal emission can be defined as the sound that is heard when air passes through nasal passage. When marked intranasal resistance to air flow is present, the speech sound may be accompanied by extra turbulent noises, called as nasal turbulence (McWilliams, 1982). It must be stressed that nasal turbulence is a severe form of audible nasal escape and that the noise generated is distracting to listeners. The extent of the velopharyngeal opening associated with audible nasal emission may vary from less than 5 mm square (McWilliams & Philips, 1989) to completely open.

Curtis (1968) and House & Stevens (1956) showed that nasal quality is characterized by a decrease in the intensity of vowels because of an increase in the absorption of acoustic energy. Audible nasal emission may well occur in cases of marginal velopharyngeal inadequacy when there is sufficient closure to generate intraoral air pressure for speech but inadequate closure to prevent nasal escape of air.

Hairfield et al., (1988) reported that 68% of 85 children and adults with cleft lip and palates were oral or mixed oral-nasal breathers, whereas only 32% were nasal or predominantly nasal breathers. The degree to which hypernasality is perceived by a listener will depend on the characteristics of the entire vocal tract and not only on the size of the functional opening in the velopharyngeal valve (Curtis, 1968).

In a study of early versus late primary hard palatal closure, Henningson, Karling & Larson (1990) reported that hypernasality was significantly worse for the late closure group than for the early closure group. Although eight of 24 (33%) were rated as 2 or higher on a scale where 0 = no hypernasality and 4 = severe hypernasality, 19 of 21 (90%) of the late closure group was rated as 2 or higher. These findings clearly demonstrate that late palate closure tends to result in greater severity of velopharyngeal inadequacy.

Fox DR, Lynch & Cronin (1988) studied on changes in nasal resonance over time. They reported that the fifteen of the 20 patients had been judged by a speech pathologist and two surgeons to have poor resonance following surgical treatment.

Van Lierda, Van Borsel et al., (2002) studied on effect of cleft type on overall speech intelligibility and resonance. The aim of the study was to measure and compare the effect of a unilateral cleft lip and palate and bilateral cleft lip and palate on overall speech intelligibility, nasalance and nasality and also to compare the nasalance and nasality patterns of cleft palate children with the available normative data for normal children. The subjects were 37 children with unilateral cleft lip and palate (19/37) or a bilateral cleft lip and palate (18/37) with normal cognitive and motor functioning and normal hearing level. A perceptual evaluation of speech, the Gutzmann (1913) test and the test of Bzoch (1989) were used as subjective assessment techniques. The findings of their study show no statistically significant differences for nasalance values, nasality and overall speech intelligibility between the UCLP and BCLP children. Significant differences were found between the data of the normal group and the cleft palate

children. These differences included nasalance values as well as nasality data and overall speech intelligibility.

Dutevall, Ejnell & Bake (2002) studied on the relationship between perceptual evaluation of speech variables related to velopharyngeal function and the patterns of nasal airflow during the velopharyngeal closing phase in speech in children with and without cleft palate. Fourteen children with cleft lip and palate or cleft palate only and 15 controls aged 7 and 10 years and were Swedish native speakers. The results were found between ratings of "velopharyngeal function" and "hypernasality" and the pattern of nasal airflow during the bilabial nasal-to-stop combination /mp/. Both the sensitivity and specificity were 1.00 for the bilabial temporal airflow measure in relation to ratings of "velopharyngeal function." The nasal airflow rate during /p/ in /mp/ had a sensitivity of 1.00 and specificity of 0.92 to 0.96 in relation to ratings of "hypernasality."

II. b. Language disorder

The child with a cleft palate does not belong to a homogenous group, and is not exempt from other communication problems. Like all children he may be subject to language delay from a range of possible causes. For example, hearing loss, if present, will slow the development of language comprehension and in consequence expressive language will also be delayed. Periods of hospitalization may interrupt periods of normal experience, so vital for language learning. Furthermore, if a child's speech efforts are not easily understood it will be difficult for parents to reinforce communication efforts. Parents may be prevented from expanding utterances because they cannot understand the child's intended

meaning, consequently usual language- stimulating opportunities may be curtailed.

Clifford (1979) pointed out that scant attention has been paid to language in children with cleft palates. Shames & Rubin (1979) suggested that "the shortcomings of research on language in cleft palate children are directly traceable to the shortcomings of language research in language."

Early studies reported that children with clefts demonstrate poorer receptive and expressive vocabulary skills (Morris, 1962; Nation, 1970a 1970b; Spriestersbach, Darley & Morris, 1958) than noncleft children.

Brennan & Cullinan (1974) compared 14 children with clefts and 14 noncleft children at a mean age of 8 years 10 months and 8 years 11 months respectively, on task involving object identification and naming. The children with cleft performed less well than did their controls. Other linguistic deficits, including a shorter mean length of utterance and a reduction in both structural complexity and the variety of words used, have also been reported for children with cleft palate (Horn, 1972; Morris, 1962).

Whitecomb, Oschner & Wayte (1976) explored the language functioning of eight children with cleft and eight normal children between five and six years of age. The children with cleft, although they had normal hearing and intelligence, were less competent on both the Developmental Sentence Scoring and the Length-Complexity Index (Shriner & Sherman 1967). Discrepancies between these two

small groups were obvious on both instruments but were more marked on the Length-Complexity Index.

Scherer & D'Antonio (1995) compared the language performance of 30 toddlers with cleft palate in the age range of 16 to 30 months with that of 30 age-matched non-cleft toddlers on four different measures: the MacArthur Communicative Development Inventory, the Preschool Language Scale, the Rossetti Infant-Toddler Language Scale, and a conversational language sample. The two groups performed comparably on the receptive language subtest of Preschool Language Scale; however, differences were evident in expressive language performance across the other language measures. The cleft group used fewer total words and fewer different words than the non-cleft group.

Scherer (1995) conducted a longitudinal investigation of early language development in six toddlers (four with cleft lip and palate and two with cleft palate only). The subjects were studied at 20, 24, and 30 months of age. The children with cleft lip and palate achieved higher standard scores on the Bayley Scales of Infant Development and the Preschool Language Scale than the children with cleft palate only. Scherer (1995) concluded that although children with cleft lip and palate demonstrate "specific deficits in language form." Children with cleft palate only showed pervasive delays in receptive and expressive language development. The delays noted in language functioning did not diminish from 20 to 30 months.

Chapman, Graham & Gooch (1998) examined the conversational skills of pre-school and school aged children with cleft lip and palate. Participants were 20

children with unilateral cleft lip and palate (10 preschoolers with the age-range of 3.7 to 4.9 years and 10 school-age children with the age-range of 7.6 to 9.6 years) recruited from the Craniofacial Team at Rainbow Babies and Children's Hospital, Cleveland (Ohio, USA) and 20 noncleft peers matched for gender, age, and socioeconomic status. Separate comparisons were made for the preschool children with cleft lip and palate and their noncleft peers, and the school-age children with cleft lip and palate and their noncleft peers on eight measures of conversational assertiveness/responsiveness and the standardized tests of pragmatics. Next, each child with cleft lip and palate was classified for level of conversational participation. Their results revealed no significant differences between the preschool and school-age children with cleft lip and palate and their noncleft peers in level of conversational participation. However, individual child comparisons revealed less assertive profiles of conversational participation for 50% of the preschool and 20% of the school-age children with cleft lip and palate.

Morris & Ozanne (2003) evaluated the language, phonetic, and phonological skills at age 3 years of two groups of young children with a cleft palate, with different expressive language proficiency at 2 years of age. Twenty children with cleft palate were allocated to two groups dependent on expressive language abilities at age 2 years. One group had normal language development and the second group had been identified as having significantly delayed (8 to 12 months' delay) expressive language development. The results indicated that the group differences were found on both language and speech abilities at age 3 years. Significant group differences were found in expressive language, percentage of consonants correct, phonetic inventory, and phonological process usage. The

later childhood or teenage years. The musculature of velopharyngeal system is intimately tied to that of Eustachian tube. As we know, the Eustachian tube provides for (1) aeration of the middle ear cavity, (2) equalization of pressure between the middle ear cavity and the ambient or atmospheric pressure, and (3) drainage of middle ear fluid and secretions into the nasopharynx. The primary muscle responsible for opening the Eustachian tube is the tensor veli palatine, the inferior bundle of which is termed the "dilator tubae." The high susceptibility of children with clefts to middle ear disease is that the cleft interferes with the ability of the tensor to open the Eustachian tube (Bluestone, 1971; Moore & Yonkers, 1986). When the Eustachian cannot open efficiently and the middle ear cannot be ventilated, the tympanic membrane retracts and an effusion from the middle ear mucosa develops as the negative pressure increases within the middle ear cavity. The negative middle ear pressure also draws bacteria up through the tube. Eustachian tube "obstruction" is seen as the primary cause for the ear disease in children with clefts, but abnormal compliance or 'floppiness" of the tube is also a factor (Bluestone, 1971; Bluestone, Klein & Kenna, 1990a). Another consequence of middle ear disease is decreased pneumatization of the temporal bone, a finding that has been reported in children with cleft palate (Stool & Winn, 1969) and in the subjects without cleft palate (Stool, 1989).

The hearing loss associated with chronic otitis media is not always exclusively conductive in nature. There have been reports of high frequency loss (Ahonen & McDermott, 1984; Manning et al., 1994). Ahonen & McDermott (1984) tested the frequency range of 250 through 20 kHz in children with repaired cleft palate and a history of otitis media compared with controlled subjects

without clefts or a history of ear diseases. The children in the cleft group had poorer hearing throughout the tested range, but statistically significant differences were found only in range above 9 kHz.

McDermott, Fausti & Frey (1986) compared children with clefts and a history of ear disease with control subjects without cleft but a high incidence of middle ear disease and found that both group showed losses in the 8 kHz to 20 kHz range that were not found in controls. They concluded that middle ear disease alone is a "sufficient condition" for loss of auditory sensitivity in the high frequencies.

Manning et al., (1994) interpreted elevated thresholds by ABR testing at 2 KHz and above as evidence of sensorineural hearing loss in two infants with clefts. In a relatively recent report (Handzic-Cuk et al., 1996), the authors stated, "Children with bilateral or unilateral cleft lip and palate and isolated cleft palate mostly suffer from moderate (21-40 db) and severe (>40 db) conductive, bilateral hearing loss by the age of 6 years, accompanied by sensorineural hearing loss of 30 db on average".

Tuncbilek, Belgin & Ozgur (2003) evaluated the otologic and audiologic status of 50 children (29 boys and 21 girls with the age range of 6-20 years) with repaired cleft lip, cleft palate, or both in Hacettepe University, Ankara, Turkey. Audiometric and tympanometric evaluation of 100 ears in 50 children were performed. Hearing levels ≤ 15 dB and middle ear pressures between -50 to +50 decaPascals were considered to be normal. Results were examined according to cleft type and laterality. The least and most affected frequencies were

calculated. A simple evaluation of speech characteristics including nasal resonance, nasal air escape, and errors of articulation was also performed. Results indicated that sixty-three of the 100 ears had normal hearing status, whereas 40 had normal middle ear pressures. No evidence was found to suggest that individual cleft type and laterality of the ear had any effect on hearing loss or middle ear disease. Two-thirds of the patients had normal or acceptable degree of language skills

Minardi, Netto, Cruz et al, (2004) investigated the auditory abilities in children with cleft lip and/or palate population. 100 children between 7 and 12 years old with cleft and 65 between 6 and 10 years old without it were taken. Conventional audiological evaluations such as pure tone, word discrimination audiometry and acoustic immittance measures. A questionnaire (Fisher) was given to parents, with 25 items about behaviors found in children with APD. Results indicated that 100% of children with cleft lip/palate showed some indicative behaviors of APD.

Chu & McPherson (2005) reviewed the audiological and otoscopic status of Chinese children and young adults seen at a cleft lip and palate clinic in Hong Kong and compare with previous studies of Western clinical populations. The hearing health status of the patients was analyzed on the basis of the results of their otoscopic, pure tone audiometric, and tympanometric examinations. The patients' sex, age, race, and type of cleft pathology were examined for their potential relationship to hearing health status. The records of 180 predominantly Chinese patients attending the Cleft Lip and Palate Centre, Prince Philip Dental Hospital/University of Hong Kong from July 1977 to December 1999 were

reviewed. The results indicated 13.4% of patient ears failed the audiometric screening and 23.7% of patient ears yielded abnormal tympanometric results. Patients' sex, age, and type of cleft pathology were found not to be related to hearing screening results. The prevalence of hearing disorder was in contrast with Western studies that show a much higher rate of audiological problems in children and young adults with cleft lip/palate. Racial factors were considered to be possible reasons for this disparity.

b. Otitis media

Otitis media means that there is an inflammation of the middle ear. Otitis media with effusion is inflammation accompanied by a collection of liquid with no perforation of the tympanic membrane. When the effusion is serous, the liquid is thin and watery. If it is mucoid, it is thick and mucous like. It is pus like when it is purulent. When the ear disease is accompanied by discharge, the condition is called otorrhea.

Gutzmann (1983) the first to recognize the increased prevalence of middle ear disease in patients with cleft palate. Paradise & Bluestone (1974) reported that occurrence of otitis media is less frequent in infants after palatal repair, and other studies (Paradise et al., 1969) have shown that hearing losses are less apparent in older children than in younger ones (Goetzinger, 1960; Graham, 1964).

Paradise, Bluestone & Felder (1969) and Stool & Randall (1967) reported that the percentage of children with fluid present in the middle ear was more than 90%. These findings motivated a major campaign for myringotomies and

placement of ventilating tubes in babies with clefts (Bluestone et al., 1972; Stool, 1989).

In a cross-sectional study of the ear status of three- year old children with repaired clefts who had not all had aggressive ear care, Rynnnel-Dagoo et al., (1992) reported that one-third had long standing secretory otitis media and that 12 of the 44 children (27%) had two or more episodes of acute otitis media. About one third had placement of ventilating tubes, but by the age of 3 years only one child still had tubes. Hearing was normal in 82% of children, and the specific antipneumococcal antibody activity was compatible with the activity found in healthy age-matched controls.

Broen, Moller & Keenan et al., (1996) studied on the comparison of hearing histories of children with and without cleft palate. Aggressive otologic management has been recommended for children with cleft palate because of the almost universal occurrence of otitis media with effusion in these children and the association of with hearing loss and In this study, 28 children with cleft palate and 29 noncleft children were seen at 3-month intervals from 9 to 30 months to compare otologic treatment and management. Hearing and middle ear function were tested at each session; information on ventilation tube placement was obtained from medical records. Ventilation tubes were placed earlier and more often in children with cleft palate, but children with cleft palates failed the hearing screening more often. The correlation between age at first tube placement and frequency of hearing screening failures was significant for the children with cleft palate, indicating that the later tubes were first placed, the poorer the child's hearing.

Liu, Sun, Wu et al., (2004) studied on to explore the effect of eardrum tubing in the repair of cleft palate on alleviating the otitis media with effusion and hearing loss in cleft palate patients. Nineteen ears of 19 cleft palate children with otitis media with effusion and hearing loss were treated with the ventilation tube insertion in the repair of the cleft palates, while the untreated opposite ears were selected as the control group. All patients were followed up from 2 weeks to 18 months postoperatively and their middle ear condition and hearing thresholds were reevaluated by otoscopy and pure-tone audiometry. Results indicated that the significant differences were found in the incidences of hearing loss between pre and postoperative patients in treated ears, and there are no differences in the untreated ears. Postoperative hearing thresholds become lower than that before the operation and no serious complications were found in the treated ears.

c. Mental retardation

Mental development is often a major issue both in the diagnoses and the outcome of major craniofacial anomalies. Nearly half of the syndromes (50 out of 100) listed by Siegel-Sadewitz & Shprintzen (1982) are reported to be frequently or probably associated with cognitive disorder. Some syndromes are associated with malformation of the brain resulting in mental retardation in 100% of affected children. Some syndromes are invariably associated with some degree of mental retardation and others are not.

Strauss & Broder (1993) studied the compare a subpopulation of persons with cleft lip/palate who have mental retardation (n = 56) to those with normal learning (n = 420), at a large university-based cleft-craniofacial center. Many of

the patients identified as having mental retardation in this sample have the diagnosis of isolated cleft palate (46.8%). Nearly half (46.3%) of the patients with mental retardation were found to have multiple anomalies, syndromes or associated medical findings. Common findings included cardiopulmonary defects, seizures, and deviations in head size. In this clinic population, mental retardation was found more commonly among African-American patients with clefts, than among Caucasian patients with clefts. Higher rates of facial disfiguration and impaired speech were found in patients with clefts and mental retardation. This research demonstrates that among a population of persons with cleft lip and/or cleft palate, there is a subpopulation of children who also have mental retardation.

Chen, Chen & Zhang (2001) investigated the possible factors involved in intelligent development of children with cleft lip and palate in china. The intelligence quotient (IQ) and developmental quotient (DQ) of 152 children with cleft lip and palate under 14 years old were conducted, and the control group was made up of 80 healthy children. The considered factors included degrees of deformation, situation of perinatal period and way of feeding, psychological conditions of the children, general situation of their parents and education of these children. They concluded that the intelligent development of children with cleft lip and palate is affected by many factors, including types and degrees of deformity, feeding ways, illnesses and social psychological factors in different periods of development.

d. Dental problem

The patients with cleft lip and palate have a significant dental problem. Individual teeth in the area of the cleft may be missing or malpositioned. The upper or maxillary arch may not fit well with the lower or mandibular arch, which is usually to as a malocclusion. Dental eruption may be delayed in individuals with clefts but no one has derived a unified theory for why the dental eruption should be differentially affected by gender or by cleft- type subgroups. (Jordon, Neptune, 1966; Ranta, 1971; Solis et al., 1998).

Ross (1975) reported that children with cleft palate alone (with no cleft of the lip or alveolus) also have a high incidence of missing permanent lateral incisors. Larson, Hellquist & Jakobsson (1998) studied on to evaluate the influence of cleft size and surgical treatment on the development of permanent teeth in patients with isolated cleft palate. The series comprised 109 children with isolated cleft palate, 70 girls and 39 boys, including 14 patients with Robin sequence. The patients were grouped according to the sagittal extent of the palatal cleft, measured on dental casts obtained before the primary palate surgery. Forty-six children were treated by one-stage palatoplasty, and 63 in two stages. The dentition was studied on orthopantomograms taken at 5, 8, 11, and 14 years of age. Congenitally missing permanent teeth (third molars excluded) were found in 33 subjects (30%). Children with large clefts had significantly more missing teeth than children with small clefts. The tooth most usually missing was the mandibular second premolar, followed by the maxillary lateral incisor, and the upper second premolar. The incidence of dental malformation was 23%, mostly mild forms. Ectopic eruption of the upper first permanent molars was seen in 23

(45%) of the subjects with large clefts, and in 18 (31%) of those with small clefts. The surgical method did not significantly affect the direction of the eruption. There is a correlation between cleft size and hypodontia, dental deformity, and ectopic eruption. Children with Robin sequence had almost the same incidence of hypodontia, malformed teeth, and ectopic eruption as children with large clefts. There was no correlation between surgical method and ectopic eruption of the maxillary first permanent molars.

A missing lateral incisor on the side of the cleft is quite common in the permanent dentition of children with clefts. It is something of a paradox that supernumerary teeth are also common (Vargervik, 1981). In young children with clefts the spaces created by missing teeth may contribute to oral distortion of complex consonants such as sibilants and affricatives, but the effect is usually temporary. In unilateral cleft lip and palate there is often a lateral crossbite on the side of the cleft, that is, the lateral segment of the maxilla is positioned medially to its counterpart in the mandible which is termed as "arch collapse". Bilateral crossbite, meaning collapse of both arches in the maxilla, is often found in bilateral clefts but may also occur in unilateral clefts, particularly when there are congenitally missing teeth. If the premaxilla is also positioned behind the anterior mandibular teeth, this is a full crossbite or a class III malocclusion.

Class II occlusions are relatively rare in clefts, except in those conditions in which the mandible is inherently small. Many children with Pierre Robin sequence show a Class II tendency in their early dentition because the mandible is small, but in the absence of a syndromic condition the mandible in patients with Robin sequence usually grows forward. In contrast, in syndromes such as

mandibulofacial dysostosis and stickler syndrome maxillary-mandibular disproportion usually remains and may in fact become more marked with growth. Dental and occlusal problems are more likely to be causative factors in speech problems (1) when they occur in combination rather than singly, (2) when they are present during speech-learning years as opposed to later years, and (3) when they influence the spatial relationship between the tip of the tongue and the incisors (Peterson-Falzone, 1994). The literature also indicates that speech problems are fairly common when there is a restriction in the size of the palatal vault and are more apt to be found in class III occlusions compared with class II (Peterson-Falzone, 1994).

Heliovaara, Ranta & Rautio (2004) studied on dental abnormalities in permanent dentition in children with sub-mucous cleft palate. Seventy-three children with submucous cleft palate (38 girls and 35 boys), mean age 8.2 years (range 7.7-9.5), were studied retrospectively from orthopantomograms. Dental abnormalities in permanent dentition were found in 26 patients (36%). Missing teeth mainly lower 2nd premolars, upper lateral incisors, and upper 2nd premolars, were found in 12 patients (16%). Most of the patients had 1 or 2 missing teeth, 2 had 3 missing teeth. In 5 patients hypodontia was associated with another dental abnormality. Other dental abnormalities included peg-shaped lateral incisors in 7 patients (10%), ectopic eruption of upper 1st molars in 6 patients (8%), transposition of upper canines and 1st premolars in 3 patients (4%), supernumerary teeth in 2 patients (3%) and palatally displaced upper canines in 1 patient (1%). As children with submucous cleft palate have a tendency towards

increased frequency of missing teeth and other dental abnormalities, the need for thorough clinical and radiological dental examination is emphasized.

Duque, Dalben & Costa et al., (2004) investigated the chronology and sequence of eruption of deciduous teeth eruption in children with complete unilateral cleft lip and palate. A total of 435 children aged 0 to 48 months who presented with complete unilateral cleft lip and palate. Results indicated that the all teeth on the cleft side in both jaws for both sexes presented a higher mean age of eruption than their homologues at the noncleft side. This difference was statistically significant for the maxillary lateral incisor, maxillary cuspid, and mandibular lateral incisor. There was a statistically significant sex difference regarding the mean age of eruption only for the maxillary second molar for the girls and mandibular cuspid for the boys. The maxillary lateral incisor on the cleft side was the last tooth to erupt, thus modifying the sequence of eruption of the deciduous teeth.

e. Syndromes Associated with Cleft lip and Palate

The term "syndrome" is used only when multiple structural defects cannot be explained on the basis of a single defect in the formation of embryonic tissue but rather appear to be the consequence of multiple defects in one or more tissue, although each of the independent defects is thought to be due to single cause (a gene defect, chromosomal abnormality, or teratogen). Examples of syndromes affecting the craniofacial complex include Van-der-Woude syndrome, Apert syndrome, mandibulofacial dysostosis, also called Treacher Collins syndrome and many more. The term "sequence" is used if all the anomalies can be explained on

the basis of a single structure or mechanical problem leading to a "cascade" of subsequent defects.

Jones (1988) reported that over 30% of 428 consecutive patients from the cleft palate program at children's hospital in San Diego had multiple anomaly syndromes. Data showed that certain speech disorders in children with clefts may be syndrome specific (Golding-Kushner, 1991). For example, the pervasive use of glottal stop substitutions has been found to be a nearly universal finding in children with Velo- cardio-facial syndrome (the most common syndrome of clefting), but less common in children with isolated clefts or other syndromes (Golding-Kushner, 1991).

Cohen & Bankier (1991) estimated that there were more than 340 syndromes involving orofacial clefting. Gorlin (1993) reported that only 5% of clefts were syndromic.

Lilius (1992) studied on associated anomalies and syndromes in Finland. He evaluated Probands with clefts born during an 11-year period, 1975-85. 1,586 probands were found of whom 345 (171 males and 174 females, 21.8%) had an associated anomaly. More male patients had cleft lips, with or without cleft palate (CL (P)) and more female patients had cleft palates (CP). The anomalies were subdivided according to anatomical site, and the biggest category was that of the extremities (29.7%) followed by cardiovascular (14.8%) and other facial anomalies (13.0%). The smallest category was chromosomal anomalies (2.7%) followed by miscellaneous anomalies (4.1%). A total of 560 malformations were found. Most anomalies per proband with clefts were found in the bilateral cleft lip

and palate subgroup (mean 1.04). The lowest (0.14) was found in the subgroup with the least severe cleft deformity, the cleft lip with or without cleft alveolus. Results indicated that there was no difference in parental age between probands with an associated anomaly and those with a solitary cleft. Anomalies were more than three times as frequent among probands with clefts as among the general population.

Onofre, Brosco & Taga (1997) studied on to evaluate the relationship between fistulae of the lower lip and cleft lip and/or palate in patients with Van-der-Woude syndrome. The medical records of 11,000 patients with cleft lip and/or palate registered at the Cleft Lip-Palate Research and Rehabilitation Hospital, University of Sao Paulo, Bauru were reviewed. Of these patients, 133 (1.2%) presented with Van-der-Woude syndrome. Results indicated that of the 133 patients, 88 (66.2%) exhibited full clefts, 22 (16.5%) only cleft lip and 23 (17.3%) only cleft palate. The lower-lip fistulae observed in these 133 patients were bilateral symmetric in 66 (49.7%), bilateral asymmetric in 42 (31.6%), microform in 19 (14.3%), median in 5 (3.8%), and unilateral in 1 (0.7%).

Marques, Barbieri & Bettiol (1998) investigated the etiopathogenesis of isolated Robin sequence. This study was carried out at the Hospital de Reabilitacao de Anomalias Craniofaciais (formerly the Hospital for Research and Rehabilitation of Cleft Lip/Palate), University of Sao Paulo, Bauru, SP, Brazil, which provides care for patients with lip/palate lesions throughout Brazil. Forty-three children were initially included in the study, seven of whom were later found to be cases of Stickler syndrome and excluded. The remaining 36 children presenting only the anomaly triad of microretrognathia, glossoptosis, and cleft

palate were followed up from the first month of life to 4 years of age with repetitive clinical and ophthalmological examination. Results indicated that a family history of cleft lip/palate was observed in 27.7% of cases, one case having a younger brother with only cleft palate but no microretrognathia or glossoptosis. Six cases of isolated cleft palate and three cases of cleft lip with or without cleft palate were present in distant relatives. Complete U-shaped cleft palate (wide cleft) was the most frequent type of cleft, which was present in 75% of cases. Only one case of incomplete cleft palate was observed, but U-shaped; 25% of the patients presented complete V-shaped cleft palate.

Murthy, Seshadri & Hussain et al., (2004) studied a case of cleft lip and palate associated with Seckel syndrome. They reported that an unusual association of complete cleft of the primary and secondary palate with Seckel syndrome. Seckel syndrome is a very rare syndrome, with only 60 reported cases in the medical literature. It is an autosomal recessive disorder characterized by birdlike face, intrauterine growth retardation, dwarfism, and microcephaly.

METHOD

The present study is a retrospective study, aiming to find the communication disorder and associated problems in children with unrepaired cleft lip and palate. The method used for this study is retrospective study in which fifty case files of children with unrepaired cleft lip and/or palate were analyzed.

Procedure

Data collection: In order to review the available records the following criteria were used. The available data was classified as four groups (a) cleft of the hard palate and soft palate (b) cleft lip associated with hard and soft palate (c) submucous cleft palate (d) Velopharyngeal dysfunction

Criteria used for selection of case records

- The cases who reported to AIISH with the history of unrepaired cleft lip and/or palate in the age range of 2- 10 years, registered between the 1st January 2002 to 31st December 2006 were reviewed.
- The case documents with oral cleft, which includes unilateral/bilateral, complete/incomplete, lip and/palate, submucous cleft palate, velopharyngeal incompetency alone, and repaired/unrepaired were considered.
- Each case file was separately analyzed in detail for the demographic information and to find the presence or absence of factors like consanguinity, family history, types of cleft, and communication disorder and other associated problems.

The parameters considered for data collection of case files were based on the demographic information such as type of cleft (cleft of the hard palate and soft palate, cleft of the lip, hard palate and soft palate, submucous cleft palate and velopharyngeal incompetency alone), gender distribution, age at first consultation, consanguinity, family history, communication problems like articulation disorder, voice disorder, resonance disorder and language disorder in terms of receptive and expressive and other associated problems like hearing loss, otitis media, mental retardation, dental problem and syndromes associated with cleft lip and/or palate.

In order to collect the data from the case files, a data sheet (Appendix I) was prepared in SPSS 10.0 version in which all the variables were entered. A numerical value was assigned to each variable for the presence or absence of the problem. The data was extracted from the case files and fed to this program.

Statistical analysis

The data was analyzed for children with cleft lip and/or palate separately using descriptive statistical analysis for many variables like family history, consanguinity, and gender distribution. To find the association between two variables cross tabulation was done followed by like Chi-Square tests, Kappa coefficient. Cramer's V was also used to know the frequency or measures of agreement or association between types of clefts and communication disorders (articulation, voice, resonance and language), associated problems (hearing loss, otitis media, mental retardation, dental problem and syndromes associated with cleft of the lip/palate). The results are presented in the form of tables representation and pie-chart.

RESULTS AND DISCUSSION

The present study is a retrospective study involving 50 case files that reported with a complaint of unrepaired cleft lip and palate between the age ranges of 2-10 years seen during the period of 1st January 2002 to 31st December 2005. The results and discussions are explained in the following headings:

I. Incidence of Communication Disorders across Groups

(1) To find the incidence of language disorder across groups

The data was analyzed in terms of the presence of receptive and expressive language disorders in four groups. The standardized language test, which assesses the receptive and expressive ability had been administered to these children depending on the age and the language. The results were compared with the available standardized norms established for that test. These available results were extracted from these files for the presence or absence of the problems related to receptive ability and the expressive ability.

(a) *Receptive language disorder:*

Group	Normal Receptive Language	Receptive Language disorder	Total
HP+SP	18	8	26
L+HP+SP	13	5	18
SMCP	1	3	4
VPD (Alone)	2	0	2
Total	34	16	50

Table 1: Distribution of receptive language ability across each group.

The above Table 1 shows the receptive language disorder across four groups. In general, the results indicate that, the disorder related to receptive ability was seen in 16 cases, out of which 8 cases (50%) were found in cleft of the hard palate and soft palate and 5 cases (31%) found in cleft of the lip, hard palate and soft palate and 3 cases found in submucous cleft palate. The normal receptive ability was observed in 34 cases, out of which 18 cases (53%) of hard palate and soft palate, 13 cases (38%) in cleft of the lip associated with cleft of the hard palate and soft palate, 1 case (3%) in SMCP and 2 cases in velopharyngeal dysfunction group.

The results clearly indicate that the receptive ability in majority of the cases was reported to be normal. This study contradicts the studies reported that children with clefts demonstrate poorer receptive and expressive vocabulary skills (Morris, 1962; Nation, 1970a 1970b; Spriestersbach, Darley & Morris, 1958) than non-cleft children. Since majority of the children had normal cognitive ability, normal hearing, the receptive ability was reported to be normal.

(b) *Expressive Language Disorder:*

Group	Normal Expressive Language	Expressive Language disorder	Total
HP+SP	17	9	26
L+HP+SP	10	8	18
SMCP	1	3	4
VPD (Alone)	2	0	2
Total	30	20	50

Table 2: Distribution of expressive language disorder across each group.

Table-2 depicts the incidence of expressive language disorder was seen in children with different groups of cleft lip and/or palate. The expressive language disorders are found in 20 cases of cleft, out of which 9 cases (45%) were seen in the cleft of the hard palate and soft palate, 8 cases (40%) of the cleft of the lip associated with, hard palate and soft palate and 3 cases (15%) with submucous cleft palate. Subjects with velopharyngeal dysfunction did not exhibit the problem and the normal expressive language ability was observed in 30 children, out of which 17 children (57%) in cleft of the hard palate and soft palate, 10 cases (33%) in cleft of the lip associated with hard, and soft palate, one case (3%) in submucous cleft and two cases in velopharyngeal dysfunction.

Receptive Language Disorder	Expressive Language Disorder		Total
	Present	Absent	
Present	16	—	16
Absent	4	30	34
Total	20	30	50

Table-3: Cross tabulation of the expressive and receptive ability

In order to find the presence of expressive language disorder in the presence of receptive language disorder, a measure of agreement between expressive and receptive language disorder was analyzed using kappa coefficient. Table 3 gives cross tabulation of the expressive and receptive ability. The results indicated that there is a significant agreement $\kappa=82.8\%$, ($p < 0.001$) between the two i.e., there is approximately 82% agreement between expressive and receptive language disorder in children with cleft lip and/or palate.

Group	Receptive Language	Expressive Language Disorder		Total
		Present	Absent	
HP+SP	Present	8	~	8
	Absent	1	17	18
	Total	9	17	26
L+HP+SP	Present	5	—	5
	Absent	3	10	13
	Total	8	10	18
SMCP	Present	3	-	3
	Absent	~	1	1
	Total	3	1	4
VPD (Alone)	Absent	--	2	2
	Total	—	2	2

Table-4: Distribution of expressive and receptive language disorder

Table 4 depicts the group- wise cross tabulation between receptive and expressive ability. A measure of agreement between receptive and expressive language disorder was calculated with kappa coefficient in HP+ SP and L+HP+SP groups. High agreement was observed in HP+SP group (kappa = 91%, $p < 0.001$) followed by cleft of L+ HP+ SP (kappa = 65%, $p < 0.01$) and other two groups were not considered because of the sample size is less.

The results of the present study supports the findings of Morris (1962); Nation (1970a, 1970b), Spriestersbach, Darley & Morris, (1958) who reported that children with clefts demonstrate poorer receptive and expressive vocabulary skills than non cleft children,. Scherer (1995) concluded that although children with cleft lip and palate demonstrate "specific deficits in language form." Children with cleft palate only showed pervasive delays in receptive and expressive language development. So, this present study also supports that there is an agreement between expressive and receptive language disorder in children with cleft of the

hard palate and soft palate (HP+SP), cleft of the lip, hard palate, and soft palate (L+HP+SP) and submucous cleft palate. Children with cleft lip and palate may show a less assertive style of conversational participation during preschool years. This was observed clearly in this study.

The result of the present study shows that, the expressive ability is poor in children with oral clefts compared to receptive ability. Children with submucous cleft and VPD did not exhibit major problem in receptive and expressive abilities.

But, the present study doesn't support the findings of Chapman, Graham & Gooch (1998), which reported that there is no expressive language deficit seen across each type of cleft. Their results revealed no significant differences between the preschool and school age children with cleft lip and palate and their noncleft peers in level of conversational participation. Since cleft lip and palate group is a heterogenous group, the results vary according to the subjects selected for the study depending on the extent of associated problem. But this study highly warns the speech pathologists for the presence of language disorder in this clinical group. A subgroup of children with cleft lip and palate might be observed as this group is at high risk for the disorder compared to children with normal orofacial structure. Since the language development depends on the extent of the cleft, presence of cognitive deficit and the other problems speech pathologist plays a vital role in prevention and management of this disorder.

II. To find the incidence of speech disorder across groups

a. Articulation Disorder

Group	Normal Articulation	Articulation disorder	Total
HP+SP	8	18	26
L+HP+SP	0	18	18
SMCP	0	4	4
VPD (Alone)	0	2	2
Total	8	42	50

Table-5: Distribution of articulation disorder across each group.

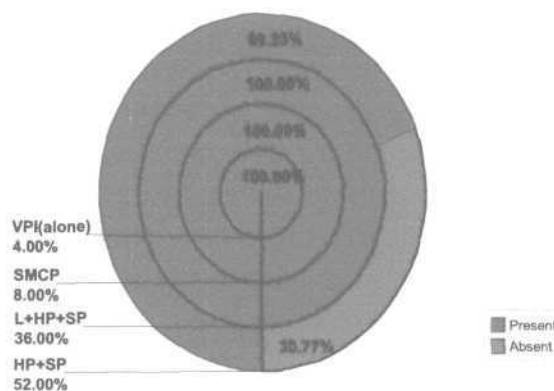


Fig 1: Clustered pie chart

Table 5 and Fig 1 represent the distribution of articulation disorder across four groups. In general, the articulation disorder was reported to be present in 42 (84%) cases and absent in 8 (16%) cases. The incidence was found to be more in children with cleft lip and/or palate and cleft lip associated with cleft of the hard palate and soft palate, 18 cases (43%) were seen in cleft of the hard palate and soft palate and also 18 cases (43%) in cleft of the lip, hard palate and soft palate, 4 cases (9%) were seen in submucous cleft palate and 2 cases (5%) in velopharyngeal dysfunction alone. To find the association between articulation

disorder and types of cleft (groups), Chi-square and Cramer's V test were used and the results indicated that there is significant association between articulation disorder and types of cleft group in Chi-square tests ($\chi^2(3) = 8.791, p < 0.05$) and from Cramer's, there is 38% association between articulation disorder and types of clefts.

This study supports the literature findings that the incidence of articulation problems is more in children with cleft palate. As Wetzel et al., (1980), Vallino & Thomson (1993) reported that formation of the sounds of speech is easily influenced by deviation in oral structures and functions so that the risk of articulation problem is high in those with cleft lip and/or palate or other cranio-facial abnormalities

Mc Williams and Musgrave (1977) & Dalston (1990) also reported that articulation abilities in children with clefts usually lag behind those of physically normal children. Seventy five percent of the younger patient in his study and 25% of the older patient demonstrated some type of communicative disorder. Articulation deficits were noted in 74 % of the 4- to 5-year-old children and in 14 % of the adolescents. Peterson-Falzone (1990) conducted a cross-sectional analysis of speech for 240 children (age 4 years to 10 years 11 months) with repaired cleft palate. More than 90 % of the young school aged children she studied demonstrated articulation problems related to place or manner of production. So, the present study supports that articulation disorder present more in cleft of the hard palate and soft palate and also in cleft of the lip, hard palate and soft palate compared to velopharyngeal dysfunction alone and submucous cleft palate.

The articulation problem is found to be more in children with oral clefts. Systematic improvement in articulation ability will be evident until approximately 10 years of age and improvement continues beyond the age but at a much slower rate. The prognosis depends on the extent of the cleft, type of the surgery, extent of the speech therapy, motivation from the client and the family members. So, the present study does support the high incidence of articulation disorder present more across the type of clefts. The incidence was found to be more in cleft associated with hard and soft palate indicating the role of the soft palate in speech production.

II. Voice Disorder

Group	Normal Voice	Voice disorder	Total
HP+SP	24	2	26
L+HP+SP	17	1	18
SMCP	3	1	4
VPD (Alone)	1	1	2
Total	45	5	50

Table-6: Distribution of voice disorder across each group.

Table 6 depicts distribution of the voice disorder seen in children with cleft lip and/ or palate across four groups. The incidence of the voice problem was found in 10% (n=5) of cases, out of which 40 % (n=2) were seen in cleft of the hard palate and soft palate (HP+SP), 20%(n=1) cases seen in cleft of the lip, hard palate and soft palate (L+HP+SP) and also followed by submucous cleft palate and velopharyngeal dysfunction group.

This findings correlate with the findings of Brooks & Shelton (1963) who reported an occurrence of 10% of voice disorder in 76 children while Takagi et al. (1965), in a retrospective study, found only 0.6% in 1061 cleft patients over a wide age range. Marks et al. (1971) also identified laryngeal dysfunction in 34% of 102. Bressmann, Sander, Horch et al., (1998) conducted a screening study for voice problem in 154 patients with cleft lip and palate patients and found that the prevalence of severe voice disorders in cleft lip and palate patients was 6.5% which is lower than reported in most other studies. Hence the present study also supports the less incidence of the voice problem compared to other speech problems.

(C) Resonance Disorder-

Resonance disorder was analysed across the four groups. The incidence of hypernasality was reported to be more compared to the voice problems. Table 7 depicts the resonance disorder presents in 43 (86%) cases in children with cleft lip and/or palate, out of which 21 cases (49%) were seen in cleft of the hard palate and soft palate (HP+SP), 16 cases (37%) in cleft of the lip, hard palate and soft palate (L+HP+SP), 4 cases (9%) in submucous cleft palate, and 2 cases (5%) in velopharyngeal dysfunction alone.

Group	Normal Resonance	Resonance disorder	Total
HP+SP	5	21	26
L+HP+SP	2	16	18
SMCP	0	4	4
VPD (Alone)	0	2	2
Total	7	43	50

Table-7: Distribution of resonance disorder across each group.

To find the measures of agreement between resonance disorder across the types of cleft (groups), Chi-square and Cramer's V test were used. The results indicated that there is no significant association between resonance disorder across groups (types of clefts) and chi-square test showed only 18% association ($\chi^2(3) = 1.692, p > 0.05$) between resonance disorder and types of clefts (groups).

The results of the study supports the findings of Van Lierda, Van Borsel et al., (2002), who studied on effect of cleft type on overall speech intelligibility and resonance. The findings show no statistically significant association for nasalance values, nasality and overall speech intelligibility between the UCLP and BCLP children. Significant differences were found between the data of the normal group and the cleft palate children. These differences included nasalance values as well as nasality data and overall speech intelligibility. So, the present study also supports that there is no statistically significant between resonance disorder and types of cleft (group).

But in a study of early versus late primary hard palatal closure, Henningsson, Karling & Larson (1990) reported that hypernasality was significantly worse for the late closure group than for the early closure group.

(II) To find the incidence of other associated problems

a. Hearing loss:

Group	Normal hearing	Hearing loss	Total
HP+SP	21	5	26
L+HP+SP	15	3	18
SMCP	3	1	4
VPD(AIone)	2	0	2
Total	41	9	50

Table-8: Distribution of hearing loss across each group.

Table 8 shows the distribution of hearing loss. The hearing loss was present in 9 (18%) cases in children with cleft lip and palate, out of which 5 cases (56%) was seen in cleft of the hard palate and soft palate (HP+SP), 3 cases (33%) in cleft of the lip, hard palate and soft palate (L+HP+SP), and 1 case (1%) seen in submucous cleft palate.

This study supports the findings of Ahonen and McDermott (1984), who tested the frequency range of 250 through 20 kHz in children with repaired cleft palate and a history of otitis media compared with controlled subjects without clefts or a history of ear diseases. The children in the cleft group had poorer hearing throughout the tested range, but statistically significant differences were found only in range above 9 kHz. Handzic-Cuk et al., (1996), stated that, "Children with bilateral or unilateral cleft lip and palate and isolated cleft palate mostly suffer from moderate (21-40 dB) and severe (>40 dB) conductive, bilateral hearing loss by the age of 6 years, accompanied by sensorineural hearing loss of 30 db on average." So, the present study supports that the hearing loss found in different types of clefts. The present study is an attempt to find the presence or absence of the hearing loss, not the extent of the hearing loss.

But Tuncbilek, Belgin & Ozgur (2003) evaluated the otologic and audiologic status of 50 children with repaired cleft lip, cleft palate, or both in Hacettepe University, Ankara, Turkey. Results indicated that sixty-three of the 100 ears had normal hearing status, whereas 40 had normal middle ear pressures.

b. Otitis media

Group	Otitis media		Total
	Present	Absent	
HP+SP	3	23	26
L+HP+SP	1	17	18
SMCP	0	4	4
VPD (Alone)	0	2	2
Total	4	46	50

Table 9: Distribution of otitis media across each group.

Table 9 depicts the incidence of the otitis media. The otitis media was seen in 4 cases (8 %) in children with cleft lip and/or palate, out of which 3 cases (75%) was seen in cleft of the hard palate and soft palate and 1 case (11%) seen in cleft of the lip, hard palate and soft palate (L+HP+SP). Forty six (92 %) children had the normal middle ear.

The present study does support the findings of Paradise & Bluestone (1974) who reported that the occurrence of otitis media is less frequent in infants after palatal repair, and other studies (Paradise et al., 1969) have also shown that hearing losses are less apparent in older children than in younger ones (Goetzinger, 1960; Graham, 1964). Rynnel-Dagoo et al., (1992) reported that one-third had long standing secretory otitis media and that 12 of the 44 children (27%) had two or more episodes of acute otitis media. About one third had placement of ventilating tubes, but by the age of 3 years only one child still had tubes. Hearing was normal in 82% of children, and the specific antipneumococcal antibody activity was compatible with the activity found in healthy age-matched controls. Rynnel-Dagoo et al., (1992) reported that one-third had long-standing

ected by many factors, including types and degrees of deformity, feeding ways, frinesses and social psychological factors in different periods of development.

(d) Syndromes associated with cleft lip and/or palate:

Table 11 shows the only 2 cases (4%) were associated with syndromes in children with cleft of the hard palate and soft palate (HP+SP). Forty eight children were ported to have non syndromic cleft. The fewer incidences of syndromes in cleft palate children were also reported by Jones (1988), who reported that over 30% of 428 consecutive patients from the cleft palate program at children's hospital in San Diego had multiple anomaly syndromes.

Group	Syndromes		Total
	Present	Absent	
HP+SP	2	24	26
L+HP+SP	0	18	18
SMCP	0	4	4
VPD (Alone)	0	2	2
Total	2	48	50

Table-11: Distribution of syndromes across each group

Onofre, Brosco & Taga (1997) studied on to evaluate the relationship between fistulae of the lower lip and cleft lip and/or palate in patients with Van de wonde syndrome. The medical records of 11,000 patients with cleft lip and/or palate registered at the Cleft Lip-Palate Research and Rehabilitation Hospital, University of Sao Paulo, Bauru were reviewed. Of these patients, 133 (1.2%) presented with Vatder Woude syndrome.

III To find the incidence of other demographic factors

(a) Gender

Group	Gender		Total
	Female	Male	
HP+SP	19	7	26
L+HP+SP	8	10	18
SMCP	4	0	4
VPD (Alone)	2	0	2
Total	33	17	50

Table-12: Distribution of gender across each group.

The above Table 12 shows the gender distribution across different groups of cleft in 50 cases of children with cleft lip and/or palate. The incidence of the cleft of the hard palate and soft palate (HP+SP) was 19 female (58%) and 7 male (41%) cases, 8 female (24%) and 10 male (59%) cases seen in cleft of the lip, hard palate and soft palate (L+HP+SP), and 4 female (12%) cases seen in submucous cleft palate (SMCP), and only 2 female (6%) cases seen in velopharyngeal in dysfunction (VPD) alone.

(b) Consanguinity:

Group	Consanguinity		Total
	Present	Absent	
HP+SP	6	20	26
L+HP+SP	5	13	18
SMCP	1	3	4
VPD (Alone)	0	2	2
Total	12	38	50

Table-13: Distribution of consanguinity across each group.

Table 13 represents the incidence of consanguinity across groups. Consanguinity was present in 12 cases (24%) in children with cleft lip and/or

palate, out of which 6 cases (50%) were seen in cleft of the hard palate and soft palate (HP+SP), 5 cases (42%) seen in cleft of lip, hard palate and soft palate (L+HP+SP), and 1 case (8%) was seen in submucous cleft palate (SMCP).

(c) *Family history:*

Group	Family History		Total
	Present	Absent	
HP+SP	3	23	26
L+HP+SP	6	12	18
SMCP	0	4	4
VPD (Alone)	0	2	2
Total	9	41	50

Table-14: Distribution of history of family across each group.

Table 14 depicts the family history across all these groups. Family history was present in 9 of children with cleft of the lip and/or palate, out of which 6 cases was seen 6 cases (67%) seen in cleft of the lip, hard palate and soft palate (L+HP+SP) and three cases (33%) were in cleft of the hard palate and soft palate (HP+SP).

SUMMARY AND CONCLUSION

Cleft is a failure of fusion of the medial nasal, lateral nasal and maxillary processes on one or both side. Cleft lip is a congenital deformity of upper lip, which varies from a notching to a complete division of the lip. It may be unilateral, bilateral or median cleft. It may be unilateral, bilateral or complete cleft palate. Cleft lip with or without cleft palate is the most common oro-facial anomaly in newborn infants, affecting one in every 750 live births.

Cleft lip and palate are associated with several problems. The type of problem depends on the extent of the cleft and other associated factors like the timing of the surgery, presence of other birth defects.etc. The associated problems may be divided into communication disorders and other problems. Communication disorders among cleft lip and palate individuals include speech disorders - articulatory disorder, voice disorder, resonatory disorders and language disorders. Other associated problems are hearing loss, dental problems, and mental retardation, with or without the presence of other syndromes.

The associated problems depend on the extent of the cleft, timing of the surgery, presence of cognitive deficit and other parameters. There are many studies done using retrospective design in order to establish the incidence of the associated problems. There are many studies done in the western context to analyze the associated problems in cleft palate population. But, there is no published study in Indian context to understand the associated problems seen in cleft palate population.

The present study is a retrospective study, aiming to find the associated problems in children with cleft lip and/or palate. Hence the method used was retrospective study and 50 case files of children with unrepaired oral cleft were analyzed.

The present study was aimed:

- (1) to find the communication disorders in children with unrepaired cleft lip and/or palate with reference to language disorder and speech disorder such as (a) articulation, (b) resonance, and (c) voice disorder
- (2) to find the other associated problems in children with cleft lip and/or palate such as hearing loss, otitis media, mental retardation, dental problem, and syndromes
- (3) find the other demographic factors such as (a) gender distribution, (b) consanguinity, (c) family history in children with cleft lip and/or palate.

The cases who reported to AIISH with the history of unrepaired cleft lip and/or palate in the age range of 2 - 10 years, registered during 1st January 2002 to 31st December 2006 were reviewed. The case documents with oral cleft, which includes unilateral/bilateral, complete/incomplete, lip and/palate, submucous cleft palate, velopharyngeal incompetency alone, and repaired/unrepaired were considered. Each case file was separately analyzed in detail to find the presence or absence of demographic factors like consanguinity, gender distribution, family history, types of cleft, and communication disorder and other associated problems. In order to collect the data from the case files, a data sheet was prepared in SPSS 10.0 version in which all the variables were entered. A numerical value was

assigned to each variable for the presence or absence of the problem. The data was extracted from the case files fed in this program. The data was analyzed for children with cleft lip and/or palate separately using descriptive statistical analysis like Chi-Square tests, Kappa coefficient and Cramer's V tests to know the frequency or measures of agreement or association between types of clefts and communication disorders (articulation, voice, resonance and language), associated problems (hearing loss, otitis media, mental retardation, dental problem and syndromes associated with cleft of the lip/palate) and other demographic factors like family history, consanguinity, gender distribution.

In general the results indicate that, the disorder related to receptive ability was seen 32% (n=16) of cases, out of which 50% (n=8) of cases found in cleft of the hard palate and soft palate (HP+SP) and 31 % (n=5) of cases found in cleft of the lip, hard palate and soft palate (L+HP+SP) and 19% (n=3) cases found in sub mucous cleft palate. The normal receptive ability was observed in 68 % of the cases, out of which 18 cases of hard palate and soft palate, 13 cases in cleft of the lip associated with cleft of the hard palate and soft palate, four cases in SMCP and two cases in velopharyngeal dysfunction group. The results clearly indicate that the receptive ability is in general reported to be normal. This study contradicts the studies reported that children with clefts demonstrate poorer receptive and expressive vocabulary skills (Morris, 1962; Nation, 1970a 1970b; Spriestersbach, Darley & Morris, 1958) than non cleft children.

The expression ability was found be normal in 40% (n=20) of cases, out of which 34.6% (n=9) of cleft of the hard palate and soft palate, 40% (n=8) of the cleft of the lip, hard palate and soft palate (L+HP+SP) and 15% (n=3) of cases

with submucuous cleft palate. Subjects with velopharyngeal dysfunction did not exhibit the problem. But the expressive disorder was observed in 30 children, out of which 17 children in cleft of the hard palate and soft palate, 10 cases in cleft of the lip associated with hard and soft palate, one case in submucuous cleft and two cases in velopharyngeal dysfunction.

The results of the present study supports the findings of Morris, (1962); Nation, (1970a, 1970b); Spriestersbach, Darley & Morris, (1958) who reported that children with clefts demonstrate poorer receptive and expressive vocabulary skills than non cleft children.. Scherer (1995) concluded that although children with cleft lip and palate demonstrate "specific deficits in language form." Children with cleft palate only showed pervasive delays in receptive and expressive language development. A subgroup of children with cleft lip and palate might be observed as this group at high risk for the disorder compared to children with normal orofacial structure

In general, the articulation disorder was reported to be present in 42 cases and absent in eight cases. The incidence was found to be more in children with cleft lip and/or palate and cleft lip associated with cleft of the hard palate and soft palate, 18 cases were seen in cleft of the hard palate and soft palate and also 18 cases in cleft of the lip, hard palate and soft palate, 4 cases were seen in sub mucus cleft palate and 2 cases in velopharyngeal dysfunction alone. This study supports the literature findings that the incidence of articulation problems is more in children with cleft palate. As Witzel et al., (1980); Vallino & Thomson, (1993) reported that formation of the sounds of speech is easily influenced by deviation

in oral structures and functions so that the risk of articulation problem is high in those with cleft lip and/or palate or other cranio-facial abnormalities.

The incidence of the voice problem was found in 10% (n=5) of case, out of which 40% (n=2) were seen in cleft of the hard palate and soft palate (HP+SP), 20% (n=1) cases seen in cleft of the lip, hard palate and soft palate (L+HP+SP) and also followed by submucous cleft palate and velopharyngeal dysfunction group. This findings correlate with the findings of Brooks and Shelton (1963) reported an occurrence of 10% of voice disorder in 76 children while Takagi et al. (1965), in a retrospective study, found only 0.6% in 1061 cleft patients over a wide age range. Marks et al., (1971) also identified laryngeal dysfunction in 34% of 102.

Resonance disorder was analyzed across the four groups. The incidence of hypernasality was reported to be more compared to the voice problems. The resonance disorder presents in 43 cases in children with cleft lip and/or palate, out of which 21 cases seen in cleft of the hard palate and soft palate (HP+SP), 16 cases in cleft of the lip, hard palate and soft palate (L+HP+SP), 4 cases in submucous cleft palate and 2 cases in velopharyngeal dysfunction alone.

The hearing loss was present in 9 cases in children with cleft lip and palate, out of which 5 cases was seen in cleft of the hard palate and soft palate (HP+SP), 3 cases in cleft of the lip, hard palate and soft palate (L+HP+SP), and 1 case seen in sub mucous cleft palate. This study supports the findings of Ahonen and McDermott (1984), who tested the frequency range of 250 through 20 kHz in children with repaired cleft palate and a history of otitis media compared with

controlled subjects without clefts or a history of ear diseases. The children in the cleft group had poorer hearing throughout the tested range, but statistically significant differences were found only in range above 9 kHz. Handzic-Cuk et al., (1996), stated that, "Children with bilateral or unilateral cleft lip and palate and isolated cleft palate mostly suffer from moderate (21-40 dB) and severe (>40 dB) conductive, bilateral hearing loss by the age of 6 years, accompanied by sensorineural hearing loss of 30 dB on average.

The otitis media was seen in 4 cases (8.0%) in children with cleft lip and/or palate, out of which 3 cases was seen in cleft of the hard palate and soft palate and 1 case seen in cleft of the lip, hard palate and soft palate (L+HP+SP). Forty six (92.0%) children had the normal middle ear The present study does support the findings of Paradise & Bluestone (1974) reported that occurrence of otitis media is less frequent in infants after palatal repair and other studies (Paradise et al., 1969) have shown that hearing losses are less apparent in older children than in younger ones (Goetzinger, 1960; Graham, 1964).

The mental retardation was seen in 6 cases of children with cleft of lip and/or palate, out of which 3 cases were seen in cleft of the hard palate and soft palate (HP+SP), 1 case seen in cleft of the lip, hard palate and soft palate (L+HP+SP) and 2 cases seen in submucous cleft palate. The study supports the less incidence of the mental retardation and does not support the findings of Siegel-Sadewitz & Shprintzen (1982), who reported that nearly half of the syndromes (50 out of 100) to be frequently or probably associated with cognitive disorder. Only 2 cases were associated with syndromes in children with cleft of the hard palate and soft palate (HP+SP). Forty eight children were reported to

have non syndromic cleft. The fewer incidences of syndromes in cleft palate children were also reported by Jones (1988), who reported that over 30% of 428 consecutive patients from the cleft palate program at children's hospital in San Diego had multiple anomaly syndromes. Onofre, Brosco & Taga (1997) studied on to evaluate the relationship between fistulae of the lower lip and cleft lip and/or palate in patients with Van der Woude syndrome.

The incidence of gender was seen more in female compared to males across each group, out of which the cleft of the hard palate and soft palate (HP+SP) was 19 in female and 7 male cases, 8 female and 10 male cases seen in cleft of the lip, hard palate and soft palate (L+HP+SP), and 4 female cases seen in submucous cleft palate (SMCP), and only 2 female cases seen in velopharyngeal dysfunction alone.

Consanguinity was present in 12 cases in children with cleft lip and/or palate, out of which 6 cases were seen in cleft of the hard palate and soft palate (HP+SP), 5 cases seen in cleft of lip, hard palate and soft palate (L+HP+SP), and 1 case was seen in submucous cleft palate.

Family history was present in nine children with cleft of the lip and/or palate, out of which 6 cases were seen in cleft of the lip, hard palate and soft palate (L+HP+SP) and three cases were in cleft of the hard palate and soft palate.

Overall the present study adds to the body of evidence that there are more communication problem seen in the articulation, resonance, and language disorder in terms of receptive and expressive disorder and less problem seen in other associated problem like hearing loss, otitis media, mental retardation and

syndromes associated with cleft lip and/or palate and also less number of cases seen in other demographic factors like consanguinity, family history but more problem seen in gender like more in female compared to male.

LIMITATIONS:

Only limited age range and limited number of case files were considered for the study.

FUTURE IMPLICATIONS.

- This study will help in the early intervention in terms medical, surgery and other modes of treatment in children with orofacial cleft, who are at risk for the development of speech and language and/ or developmental delays.
- Provides baseline information for the team members when formulating the type and intensity of prevention program needed for each child.
- Provides not only regarding therapeutic remediation rather it serves to monitor the development of a child's communication over time and to allow for initiation of therapy and/or appropriate referral as soon as a problem arises in the area of voice, resonance, phonology, language, hearing and other related factors.

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APPENDIX I

Demographic Information:

- Case number
- Age
- Gender

Groups

- Cleft of the hard palate and soft palate
- Cleft of the lip associated with cleft of hard and soft palate
- Submucous cleft
- Velopharyngeal dysfunction

Associated Communication Problems

I. Communication Disorders

a. Language disorder

- Receptive disorder
- Expressive disorder

b. Speech Disorders

- Articulation disorders
- Voice disorder
- Resonance disorder

II. Associated other Problems

- Hearing loss
- Otitis Media
- Mental Retardation
- Dental problems
- Syndromes

III. Other variables

- Family History
- Consanguinity