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A Descriptive Analysis of Individuals with Congenital Palatal Insufficiency

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A DESCRIPTIVE ANALYSIS OF INDIVIDUALS WITH CONGENITAL PALATAL INSUFFICIENCY

by

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CHAPTER I

THE REVIEW OF THE LITERATURE AND PURPOSE OF THE STUDY

Congenital palatal insufficiency (CPI) refers to an anatomical/physiological condition resulting in hypernasal speech in the absence of an overt cleft of the palate. Some authors (Lubit, 1967; Peterson-Falzone and Pruzansky, 1976) include submucous clefts of the palate (SMCP) under the general heading of CPI. A SMCP is a congenital defect characterized by an imperfect union of the velar musculature. A triad of visible stigmata typically used to determine the presence of a SMCP include the muscular disunion in the soft palate with accompanying zona pellucida, a bifid uvula, and notching of the posterior border of the hard palate. Velopharyngeal insufficiency resulting in hypernasal speech is usually evident (Calnan, 1954). Other authors (Calnan, 1954; Crikelair et al., 1970) deal with SMCP as a condition separate from CPI.

While increased attention to CPI has been evident in recent years, the identification of the condition spans more than a century. Winters (1966) identifies Passavant as the first author to acknowledge velopharyngeal insufficiency in the absence of a cleft. He contends that Passavant referred to an individual with velopharyngeal incompetency in 1862. Winters also cites Demarquay as the first individual to identify submucous cleft of the hard and soft palate in 1846. Other authors (Blackfield et al., 1962; Calnan, 1954; Dorrance, 1930; and Weatherley-White, 1972) have suggested that Roux first identified CPI in 1825, but Winters contends that Roux actually described a palatal fistula and a submucous cleft of the hard palate.

Stimson (1909) gave a lengthy description of an individual with congenital insufficiency of the palate. He describes a woman with a SMCP and hypernasality of speech. In 1892, Lermoyez (as cited in Blackfield et al., 1962) reported on twelve subjects with a condition he described as "velopalatine insufficiency." These were subjects exhibiting inadequate velopharyngeal closure in the absence of a palatal cleft. He attributed their velopharyngeal insufficiency to anatomical shortness of the hard palate. Gutzman (as cited in Blackfield et al., 1962), in 1899, suggested the possibility of deficiency of the length of the soft palate as well as the hard palate causing CPI. Kelly (1910) cited three possible causes for the condition including (1) a short palate and a deep pharyngeal vault (2) a palate of adequate length but defective in movement (3) a combination of the above. In doing so, Kelly was the first to report a physiological dimension in addition to the previously suggested anatomical causes. Dorrance (1930) cites all of the above conditions as variables affecting CPI and includes two others, SMCP and velopharyngeal insufficiency secondary to successful cleft palate surgery.

Presently, there is considerable disagreement regarding the identification of CPI. Peterson-Falzone and Pruzansky (1976) suggest two major categories which, in effect, coincide with a SMCP/velopharyngeal insufficiency separation. They identify (1) CPI with visible stigmata (2) CPI without visible stigmata. The stigmata they refer to are the soft palate muscular disunion, bifid uvula, and palatal notching characteristic of SMCP. Lubit (1967) agrees with this grouping. Calnan (1954), however, makes a clear distinction between SMCP and other causes of velopharyngeal insufficiency such as congenital short palate, cerebral

agenesis, paralysis of the palate and functional rhinolalia (i.e. nasality).

Peterson-Falzone and Pruzansky (1976) add a subgroup to their two main divisions. They classify presence or absence of a cleft lip as another anatomical consideration.

Other anatomical characteristics constituting CPI also are inconsistent in the literature. Neiman and Simpson (1975) suggest the following: (1) congenitally short velum (2) excessively deep pharynx (3) midline defects of the hard palate (4) cervical spine anomalies (5) any combination of these. Porterfield et al., (1966) add basilar skull defects to these possibilities. Others (Calnan, 1956; Lubit, 1967) include palatal paralysis as a type of CPI but generally, neurologically-based etiologies represent a separate categorization.

Although a wide variety of possible causes of CPI exists, some general conclusions can be drawn. Most authors agree that there is no overt cleft in these individuals. Velopharyngeal insufficiency signaled by nasal escape of air and hypernasal speech is a determining factor for diagnosing CPI. The neurological systems of these individuals are intact in that there is no paralysis of the velopharyngeal mechanism. Inability to close off the velopharyngeal port for anatomical and physiological reasons is present in individuals with CPI.

Management for velopharyngeal insufficiency accompanying CPI may involve surgery, speech therapy, prosthetics, or any combination of these. All procedures used are aimed at improving or correcting the speech characteristics of the individual and are generally the same as the secondary procedures used for treating velopharyngeal insuffi-

ciency following palatal surgery for clefts.

The literature points to several avenues for surgical correction of the condition. Treatment of SMCP usually involves palatoplasty to reunite velar musculature (Crikelair et al., 1970). Pushback procedures involving suturing and retropositioning of the palatal mucosa to add overall length to the palate, may be used alone or in combination with any of the following operations in the correction of palatal insufficiency (Fisher and Edgerton, 1975; Porterfield et al., 1966; Porterfield and Trabue, 1965). The pharyngeal flap is a surgical technique commonly employed in treating CPI. Its purpose is to increase velopharyngeal closure by suturing a flap of excised tissue from the posterior pharyngeal wall to the velum. Another management technique is pharyngoplasty. This is any surgical procedure that decreases the distance between the velum and the posterior pharyngeal wall by adding tissue bulk to the posterior pharyngeal wall. Retropharyngeal implants, inert substances injected into the posterior pharyngeal area to increase velopharyngeal closure, are used also. Fisher and Edgerton (1975) propose the combined use of levator retroposition (a surgical procedure designed to place the levator muscles back to an appropriate anatomical position) and pharyngeal flap. Other combinations of surgical methods also are possible.

Failure of any of these surgical methods to attain satisfactory speech results can be attributed to a host of factors. Misdiagnosis of the specific anatomical/physiological characteristics of a defective palate prior to surgery is one reason for failure. Sometimes incorrect choice or combination of surgical techniques may yield poor results.

Additionally, poor or inadequate surgical procedures can lead to less than optimal speech results. No procedure has been consistently and totally effective in alleviating velopharyngeal insufficiency.

Speech therapy is generally considered essential in the treatment of hypernasality, nasal escape of air, and articulation disorders for individuals with CPI (Porterfield et al., 1966; Porterfield and Trabue, 1965). Particular emphasis is placed on post-surgical therapy for the elimination of previously learned habits that may interfere with a newly constructed mechanism for velopharyngeal closure (Randall, Bakes & Kennedy, 1960).

Therapy techniques specific to the CPI population are not well documented. Cole (in Grabb et al., 1971) does present a method of muscle training for improving velopharyngeal function. He poses indirect, semidirect and direct forms of muscle training. Indirect muscle training is accomplished through work on the subject's articulation, since velopharyngeal closure is involved in the production of all English phonemes (except the nasal phonemes) to some degree. Yawning and swallowing exercises are incorporated at this level also. However, Cole feels that little, if any, improvement is accomplished with indirect training since new muscular activity is minimal. With the semidirect approach, increased muscular activity and improved velopharyngeal closure are emphasized through the use of techniques that do not require actual manipulation of the palatal and pharyngeal areas. Blowing and sucking exercises are employed to reach this goal. Cole's criticism of this method is that insufficient resistance often is used in semidirect exercises. No improvement in muscular activity

will occur if the subject does not encounter a large amount of pressure in the exercises performed. The final approach is direct muscle training. Cole emphasizes this last method. Direct manipulation through touching, stroking, moving, and applying resistance is used to increase awareness and control of normally involuntary motor acts. After a three month period of muscle training, evaluation of progress is suggested. No further increase in muscle activity is likely to occur after this amount of time. Pannbacker (1973) provides a comprehensive review of documented speech therapy procedures used with cleft palate speakers. After presenting information on articulation, velopharyngeal closure, and voice therapies for cleft palate individuals she concludes that therapy is often useful for functional articulation and voice problems but that surgical or prosthetic management is in order when physical deficiencies cannot be overcome. Generally, her assumptions are equally applicable to speech therapy specific to CPI subjects.

Speech therapy may fail to produce an appreciable decrease in hypernasality (and corresponding increase in velopharyngeal closure) because of an anatomical/physiological deficiency in the individual. Incorrect selection of therapeutic techniques and poor or inadequate surgical management are other causes of failure. The intelligence of the individual and other variables may contribute to failure too. No consistently effective speech therapy techniques have been documented.

Prosthetic management is another possibility in the correction of hypernasality in CPI. As Grabb et al. (1971) note, "Speech appliances may be constructed not only for persons with cleft palate but also for individuals with congenitally acquired (sic) palatal insufficiency."

(p. 886) Both palatal stimulators and palatal lifts have been suggested for use by patients displaying palatal incompetency (Grabb et al., 1971). Speech bulbs are also feasible forms of prosthetic management. With any of these devices the possibility of failure again exists. This can be the result of improper prosthetic fitting, inappropriate choice of type of prosthesis, and inability of the musculature to compensate even with the prosthesis in place. Two other cautions involved in the fitting of any oral prosthesis designed to aid velopharyngeal closure are the possible rejection of the device by the client and the effect of the device's torquing action on the existing dentition and ultimate loss of supporting dental structures (Peterson-Falzone and Pruzansky, 1976). The intellectual functioning of the individual may present additional difficulty. The relative high cost of maintaining obturators and the fact that prosthetic management is rarely if ever a lifelong solution are two additional reasons for questioning this type of treatment. Little mention of this form of management is made in the literature in reference to the CPI population.

Other factors related to the CPI exist also. Ear problems such as chronic otitis media resulting from eustachian tube malfunctioning and nasal regurgitation of liquids were found to be presenting problems in a SMCP population (Beeden, 1972). Hearing loss related to recurrent middle ear infections may be associated with this population too (Pannbacker, 1973). The time of onset of the problem may yield valuable information. A number of authors (Beeden, 1972; Calnan, 1958; Crikelair et al., 1970; Gibb, 1958; Lubit, 1967; Neiman and Simpson, 1975; Porterfield and Trabue, 1965; Randall et al., 1960; and Thaler et al., 1968) refer to individuals in whom hypernasality and inadequate velo-

pharyngeal closure become apparent following removal of tonsils and adenoids. These conditions may also become apparent as tonsils and adenoids undergo normal involution with age. Pruzansky et al. (1977) call this a lanthanic condition since it remains unidentified until some unrelated treatment is performed. Lubit (1967) contends that CPI is not diagnosed until the child begins talking. Referring to a SMCP population, Beeden (1972), Crikelair et al. (1970) and Thaler (1968) all feel that diagnosis of this condition is often made only after a problem presents itself. These remarks infer that further examination of time of onset of the problem may yield valuable information. Additionally, source of referral and original complaint can be helpful diagnostic information (Porterfield and Trabue, 1970). Genetic transmission of the condition is another possibility (Randall et al., 1960). Little information on the above mentioned variables is currently available.

The Purpose of the Study

It is the purpose of this study to provide a descriptive analysis of 20 CPI subjects examined at the Butterworth Hospital Oral Cleft Clinic between September 1974 and June 1977. Time of onset of the problem and source of clinic referral will be reported. Anatomical and physiological characteristics of the subject population will be discussed. Treatment procedures and results will be examined also with primary emphasis placed on surgical and therapeutic management of the velopharyngeal insufficiency and the accompanying hypernasality

of speech.

More specifically, this study seeks to answer the following questions:

1) What problem(s) caused the subjects to seek examination by the Oral Cleft Clinic team?
   a. What (if any) event leads to the identification of the condition in the subjects?
   b. At what age does the problem become evident?
   c. What professional (or person) refers the patient to the clinic?
2) What characteristics are present in this population of CPI subjects?
   a. What anatomical/physiological characteristics are present?
   b. What surgery/speech therapy characteristics are evident?
   c. What related characteristics are identifiable?
3) What procedures are followed with respect to surgery and speech therapy in the treatment of velopharyngeal insufficiency in this population of CPI subjects?
   a. What management technique is reported to most frequently result in improved speech?
   b. What sequence of management is followed with respect to the chronological ordering of speech therapy and surgical procedures?

CHAPTER II

PROCEDURES AND METHODS

Subject Selection

Two criteria were used in the selection of subjects for this study. First, a diagnosis of CPI by the Butterworth Hospital Oral Cleft Clinic team of Butterworth Hospital in Grand Rapids, Michigan was identified. Second, if such a diagnosis was not stated explicitly, evidence of symptomatology indicative of CPI appearing in the Oral Cleft Clinic charts was utilized. The prime symptomatic criterion was evidence of insufficient velopharyngeal closure in the absence of an overt cleft of the palate or palatal paralysis. In addition, individuals diagnosed as having SMCP, a congenitally short palate, a deep pharyngeal vault, or a high arched palate (all in the absence of an overt cleft of the palate) were included in the subject population.

A group of 51 patients examined at the Oral Cleft Clinic between September 1974 and June 1977 met the above criteria. Letters with response postcards were sent to these subjects to determine those willing to complete a questionnaire. A copy of the cover letter may be found in Appendix A. The 20 subjects responding positively were identified as the subject population.

Variables

Variables determined to have significant informative value

concerning the CPI population as evidenced by the literature included time and age of problem onset, source of clinical referral, anatomical/physiological characteristics, characteristics of management, and other related characteristics. Anatomical/physiological variables noted were (1) presence or absence of reported SMCP, bifid uvula, a notch in the posterior part of the hard palate, congenitally short palate, deep pharyngeal vault, high palatal vault, and/or cleft lip; (2) presence or absence of reported velopharyngeal closure and accompanying hypernasality of speech; (3) presence or absence of reported feeding, swallowing, sucking, and/or blowing difficulties; (4) presence or absence of reported hearing loss and/or ear infections. Characteristics of management examined were (1) reported frequency and effect of speech therapy; (2) type and result of reported surgical procedures; (3) reported use of prosthetic devices; (4) reported sequencing of management techniques. Other related variables studied were (1) presence or absence of reported articulation problems, language problems, and/or evidence of decreased intellectual functioning; (2) presence or absence of reported allergies/upper respiratory tract infections and/or mouth breathing; (3) presence or absence of a genetic/chromosomal component and/or congenital abnormalities other than CPI.

Questionnaire

A questionnaire was designed to validate and supplement information obtained from the Butterworth Hospital Oral Cleft Clinic files and to gain pertinent information about the time period following each subject's last clinic visit. Topic areas covered were identifying information,

history of the problem, surgery, speech therapy, hearing, and additional information. Specific historical considerations included source of referral, nature and onset of the problem, and speech and speech related characteristics. A listing of the time, place, and nature of surgery as well as the performing surgeon and effect of surgery on speech follows. Information concerning amount of therapy, therapeutic setting, frequency of services and group versus individual therapy was requested next. Additionally, informants were asked to provide a description of the effect of therapy on speech. Hearing specifications included a description of any known hearing loss and a listing of all otological treatment procedures used. Space for the inclusion of other significant and related information was provided also. A copy of the questionnaire and explanatory letter may be found in Appendix B.

Data Collection and Analysis

Analyses for this study were based on data collected from the Butterworth Hospital Oral Cleft Clinic files and on responses to the CPI questionnaire. Where appropriate, additional information was obtained from medical files. Information from each Oral Cleft Clinic examination, from the CPI questionnaire, and from the medical files was compiled on a master data chart (see Appendix C). This chart is divided into three main categories: (1) anatomy/physiology, (2) management, and (3) related characteristics. Anatomical features of interest were SMCP, bifid uvula, palatal notching, short palate, deep pharyngeal vault, high palatal vault, and cleft lip. Physiological

considerations included inadequate velopharyngeal closure and feeding, swallowing, sucking, and blowing difficulties. Hearing loss and evidence of ear infections are other variables in this category. The management section is comprised of speech therapy, surgery, and prosthetics. Specific surgical operations are listed. Those considered to be relevant to this study are palatoplasty, pushback procedures, pharyngeal flap, pharyngoplasty, and retropharyngeal implants. Additionally, tonsillectomy/adenoidectomy (T&A), cleft lip repair, and ear surgery were felt to be important surgical factors. The third division of the chart is composed of other related characteristics. Those listed are articulation problems, language problems, decreased intellectual functioning, allergies/upper respiratory tract infections, mouth breathing, genetic/chromosomal abnormalities, and congenital abnormalities. The presence or absence of a characteristic or utilization/non-utilization of a management technique was denoted by a plus (+) sign or a minus (-) sign respectively. A notation of "no information" (NI) or "not applicable" (NA) was made where appropriate. Information concerning nature and source of referral, original complaint, time and age of onset, treatment sequence, effect of management on speech, and other pertinent information not amenable to a dichotomous analysis was compiled separately. Frequency and descriptive analyses were conducted on the data.

CHAPTER III

RESULTS

The subject population consisted of 12 females and eight males whose ages at the time of study ranged from five years, six months to 22 years, eight months. Table 1 shows a frequency distribution of reported time of onset of the problem. In 13 cases, problems related to CPI were reported to be apparent in infancy. In two cases, the problem became more evident following the removal of tonsils and adenoids. In two other cases, onset of speech was reported to have revealed the problem. Onset time for three subjects is unknown but in two of these cases, the condition was identified by the speech therapists when these subjects were of school age. No early childhood information is available for the remaining subject.

Table 1. Frequency distribution of reported time of onset of the problem.

Reported onset time	Number of subjects	Percentage of subjects
Infancy	13	65
After tonsillectomy and/or adenoidectomy	2	10
With speech	2	10
Unknown	3	15

Table 2 lists the frequencies and corresponding percentages of persons responsible for referral of the subjects to the Oral Cleft Clinic. Speech therapists were involved in the referral of 15 subjects. Four of these were made in conjunction with other professionals, two with physicians and two with school nurses. Other sources of referral included three by plastic surgeons, one by an otolaryngologist, and one by a parent.

Table 2. Frequency distribution of source of Oral Cleft Clinic referral.

Source of referral	Number of subjects referred	Percentage of subjects referred
Speech therapist	11	55
Speech therapist and physician	2	10
Speech therapist and school nurse	2	10
Plastic surgeon	3	15
Otolaryngologist	1	5
Parent	1	5

The anatomical features studied and the reported frequency of occurrence in the subject population are found in Table 3. The anatomical conditions studied included submucous cleft of the palate (SMCP), bifid uvula, palatal notch, congenitally short palate, deep pharyngeal vault, high palatal vault, and cleft lip. SMCP accompanied

by a bifid uvula and posterior notching of the hard palate was present in six subjects. There is one report of a notch and no SMCP, resulting in a total of seven reported palatal notches. Information sources indicate that 15 subjects had congenitally short palates. Deep pharyngeal vaults were reported to be present in nine individuals with three subjects described as having high palatal vaults. Cleft lip was present in three subjects. Of the 20 subjects, 19 possess at least one of these anatomical features. Appendix C includes a breakdown of anatomical conditions for individual subjects.

Table 3. Frequency of occurrence of reported anatomical characteristics.

Anatomical Characteristic	Number of subjects presenting the characteristic	Percentage of subjects presenting the characteristic
Submucous cleft of the palate (SMCP)	6	30
Bifid uvula	6	30
Palatal notch	7	35
Congenitally short palate	15	75
Deep pharyngeal vault	9	45
High palatal vault	3	15
Cleft lip	3	15

Table 4 lists the frequency of occurrence of reported physiological characteristics including velopharyngeal insufficiency, difficulties

in related non-speech activities (i.e., feeding, swallowing, sucking, and/or blowing), hearing loss, and ear infections. There is some indication of velopharyngeal insufficiency for all subjects. The occurrence of any one of the four non-speech physiological characteristics examined is included also in Table 4. Twelve subjects were reported to have difficulties in at least one of these areas. There are no reported difficulties in six subjects and information for two individuals is unavailable. Hearing loss was reportedly present in four subjects and absent in the remaining individuals. Evidence of ear infections is reported in 11 subjects, with no ear infections indicated for eight others. Information is unavailable for one subject. A complete listing of all physiological conditions for individual subjects may be found in Appendix C.

Table 4. Frequency of occurrence of reported physiological conditions.

	Number of subjects		
Physiological condition	Condition present	Condition absent	No information
Velopharyngeal insufficiency (VPI)	20	0	0
Difficulties in related non-speech activities	12	6	2
Ear infection	11	8	1
Hearing loss	4	16	0

The reported frequency of occurrence of other selected characteristics possibly related to the CPI condition is listed in Table 5. Those conditions identified were articulation problems, language problems, evidence of decreased intelligence, allergies/upper respiratory tract infections, mouth breathing, genetic/chromosomal abnormalities, and congenital abnormalities. Articulation difficulties unrelated to resonance problems reportedly occurred in 13 subjects. Records of four subjects indicate good articulation and no information was available for three others. Positive identification of language problems was reported for six individuals. No problems were reported to be present in eight others and information on the remaining six subjects was not available. Evidence of below average intelligence was suggested in the records of eight subjects while other records indicate average or above average intelligence. Allergies and/or upper respiratory tract infections were reported for eight individuals, six subjects were reportedly free of such problems and information was unavailable for six subjects. A history of mouth breathing was present in five subjects. No mention of this condition was made for the 15 other subjects. Reported genetic/chromosomal abnormalities were present in five subjects. Eight showed no reported indication of problems in these areas. Information was not available for the remaining seven individuals but the possibility of the existence of such abnormalities was suggested for four subjects. Six subjects were reported to have congenital abnormalities other than CPI. No reported abnormalities were present for the remaining subjects. Appendix C lists the related characteristics for each member of the

population.

Table 5. Frequency distribution of reported occurrence of conditions possibly related to CPI.

Condition	Number of subjects		
	Condition present	Condition absent	No information
Articulation disorders	13	4	3
Language disorders	6	8	6
Below average intelligence	8	12	0
Allergies/upper respiratory tract infections	8	6	6
Mouth breathing	5	-	15
Genetic/chromosomal abnormalities	5	8	7
Congenital abnormalities	6	14	0

A breakdown of the type and frequency of management procedures used in treating the subjects is found in Table 6. Eighteen of the 20 subjects had received speech therapy. One subject had no speech therapy and information on early childhood therapeutic history was unavailable for another subject. Surgical operations related to the speech and/or hearing mechanism have been performed on 16 subjects. A breakdown of reports of specific surgical techniques, as seen in Table 6, indicates that six subjects had palatoplasty in conjunction with a superiorly based pharyngeal flap. Two retropharyngeal implants and

one pharyngoplasty were performed. Twelve subjects had tonsillectomies and/or adenoidectomies. All three subjects with cleft lip had surgical repair of the lip. Ear surgery was performed on seven of the 20 subjects and included a myryngotomy and/or the insertion of polyethylene tubes. Neither pushback operations nor oral prostheses were used in the management of this subject population. Refer to Appendix C for a listing of individual treatment procedures employed.

Table 6. A list of management procedures and their frequency of use on the subject population.

Management Procedure	Number of subjects		
	Procedure utilized	Procedure not utilized	No information
Speech therapy	18	1	1
Palatoplasty	6	14	0
Pharyngeal flap	6	14	0
Retropharyngeal implant	2	18	0
Pharyngoplasty	1	19	0
Tonsillectomy and/or adenoidectomy (T&A)	12	8	0
Lip repair	3	17	0
Ear surgery	7	13	0
Palatal pushback	0	20	0
Prosthetics	0	20	0

Reported effects of the management procedures on the speech characteristics of the subjects are summarized in Table 7. Of the 18 subjects known to have received speech therapy, speech was reported to have improved in 12 subjects. No change in speech was reported for the remaining six individuals. Surgical operations were reported to have positively affected five of the six subjects with combined palatoplasty and pharyngeal flaps. The sixth subject treated by this method had no reported change in speech. Speech evaluation for this individual was conducted one week post-operatively. The speech of both individuals receiving retropharyngeal implants reportedly improved as a result of the surgery and no change was reported in the speech of the subject receiving the pharyngoplasty. Of the twelve subjects having tonsillectomies and/or adenoidectomies (T&A's), a negative effect on speech was reported in five cases, five others showed no change, and speech improvement was noted in two individuals.

Table 7. Reported effect of management procedure on speech.

	Number of subjects		
Management procedure	Improved speech	Worsened speech	No change in speech
Speech therapy	12	0	6
Palatoplasty/ pharyngeal flap	5	0	1
Retropharyngeal implant	2	0	0
Tonsillectomy and/or adenoidectomy	2	5	5
Pharyngoplasty	0	0	1

A summary of the treatment sequence is shown in Table 8. The examination of the subjects at the Oral Cleft Clinic was included in this sequencing of events since, in analyzing the data, the Oral Cleft Clinic visit appeared to be a significant landmark in the management of these individuals. For eight subjects, speech therapy was followed by the Oral Cleft Clinic visit and surgery (or recommended surgery) for velopharyngeal insufficiency. Five others followed this same order of events but received additional speech therapy postoperatively. Five of the subjects received speech therapy, were seen at the clinic, and continued with speech therapy. No surgical recommendations were made for these five individuals. One subject was referred to the Oral Cleft Clinic because of a SMCP and surgical correction of velopharyngeal insufficiency was recommended. No early childhood information is available for one subject, but it is known that this individual had a diagnostic speech evaluation followed by the Oral Cleft Clinic visit and surgical recommendation for treatment of an anterior nasal obstruction.

Table 8. Frequency distribution of reported treatment sequence.

Reported sequence	Number of subjects	Percentage of subjects
Speech therapy, Oral Cleft Clinic, surgery	8	40
Speech therapy, Oral Cleft Clinic, surgery, speech therapy	5	25
Speech therapy, Oral Cleft Clinic, speech therapy	5	25
Oral Cleft Clinic, surgery	1	5
Speech therapy examination, Oral Cleft Clinic, surgery	1	5

CHAPTER IV

DISCUSSION

The results of this study suggest four main trends regarding the onset of congenital palatal insufficiency. It is apparent that onset is associated with infancy, signaled by some obvious physical abnormality such as cleft lip or by deviations in related physiological functions such as feeding or sucking. If the problem is not evident during infancy the degree to which the condition affects speech may be another indication of onset. The speech may be severely affected and thus onset may be associated with the initiation of speech. Onset also may be associated with the removal of tonsils and adenoids. This removal produces a change in physical structures which is perceived as a problem post-surgically. Finally, the degree of severity may not interfere with speech intelligibility and no problem may be evident to the subject or to his/her parents. In such cases, the problem is not perceptually significant until the child is monitored by a professional, most often the speech therapist. These trends yield variable ages for onset time and although specific age of onset is unknown the results do imply that some evidence of the condition is present and may be identified in the first few months of life, confirming the notion that it is truly a congenital anomaly.

There is a distinction between time of onset and the actual diagnosis of an anatomical/physiological deficiency. Diagnoses are reported to be made after there is an indication of an existing problem.

Often, they are not made until the individual is seen by a team of professionals. Even in the case where an obvious physical abnormality (cleft lip) was present and treated, a structural defect (SMCP) indicative of CPI was not diagnosed until the subject was school age. This subject's mother reports: "His problem could have went (sic) unnoticed for quite a time if not for the speech therapist's interest." Another parent reported that the problem "was apparent from birth (1972) but never diagnosed till 1977." These comments suggest the need for early recognition and diagnosis of the condition.

When describing problems that result in referral to the Oral Cleft Clinic, such terms as "poor speech," "talking through the nose," and "speech difficulties" were used by the informants. The frequent use of these speech-related terms correlates with the source of the referral. Speech therapists made the largest number of referrals for this study group. This fact can lead to a number of inferences. It is possible that a physical defect such as SMCP is readily apparent to the speech therapist and, as such, warrants immediate medical referral. Also one can assume that it is the speech therapist who is trained to recognize those speech disorders that are in need of remediation in addition to, or apart from, speech therapy. As one informant states, "His (the subject's) problem wasn't noticeable to anyone who wasn't trained in that type of problem as he had no visible disorders." It appears that the speech therapist plays an important role in identifying these CPI individuals.

Results indicate individual variation among conditions studied.

No two subjects present the same anatomical/physiological, surgery/speech therapy, or related characteristics. An implication of this finding is that CPI is manifested in a variety of ways and reflects marked individual differences.

Although variations do exist, some conditions are more common than others. Anatomically, the congenitally short palate is the most frequently reported feature. Other anatomical conditions such as SMCP, bifid uvula, palatal notching, deep pharyngeal vault, high palatal vault, and cleft lip are present in at least one of the subjects but no particular pattern of occurrence is readily observable.

Physiologically, only one characteristic is present in every member of the subject population and that characteristic is velopharyngeal insufficiency (VPI), typically resulting in hypernasality. Although there is some reference to this term in the available sources of information for all subjects, inconsistencies in application of the term exist. These discrepancies appear to be attributable either to disagreement among different professionals or to the change in speech characteristics as a function of time. Differences might also be attributed to the number of terms that were used to describe this condition in the subjects. Such words as "velopharyngeal insufficiency," "hypernasality," "nasal speech," "air leakage," and "air escape" were found in the sources of information. Due to the retrospective nature of this study, no operational definition of VPI can be applied. Thus, interpretation and application of terms was left to the individual supplying the information. Only the internal definitions and the expertise of the examining professionals lend credibility to the

information supplied. Regardless of the above difficulties, VPI and accompanying hypernasality of speech are of prime importance in the description of CPI. It is the only variable of any kind to be present in all subject members.

Feeding, sucking, swallowing, and/or blowing difficulties are present in a large percentage (60%) of the subjects. Early identification of the existence of a problem often is reported in relation to the difficulties a child has in one or all of these activities. Certainly it is not possible to establish a cause/effect relationship between such difficulties and the CPI condition, but the fact that the majority of subjects showed some indication of difficulty in these areas does at least warrant concern.

Ear infections, generally considered to be characteristic of the cleft palate population, also showed a high percentage of occurrence (55%) in this CPI population. Research confirms the relationship of this variable to eustachian tube malfunctioning in cleft palate individuals. In the case of CPI, ear infections may be caused by a malpositioning of the palatal musculature anatomically near the eustachian tubes with concomitant malfunctioning. Permanent hearing loss is noted in four subjects. These results do not reflect reports of fluctuating hearing loss related to chronic middle ear infections.

Evidence of all related characteristics exists for some subjects. These characteristics were articulation problems, language problems, decreased intelligence, allergies/upper respiratory tract infections, mouth breathing, genetic/chromosomal abnormalities, and congenital abnormalities. Articulatory problems unrelated to resonance problems

shows the highest incidence of occurrence of all related variables. They are reportedly present in 60% of the population studied. This fact is especially important when considering the treatment of the subjects and will be discussed below. Each of the other related characteristics is present in at least 25% of the population. Speculatively, this frequency of occurrence is greater than the frequency of occurrence in the general population. Consequently, the need to account for these characteristics when examining for CPI should not be overlooked. It is possible that interrelationships exist among these variables and further study examining these relationships is indicated.

The management procedures used to treat the CPI population in this study were surgery and speech therapy. Prosthetic management, sometimes employed as treatment of CPI was not used in the treatment of any of these subjects.

Speech therapy was the most frequently utilized form of management with 90% of the population in this study having received speech therapy services. This therapy may or may not have been concerned with direct alleviation of the velopharyngeal insufficiency secondary to CPI. As noted earlier, direct, semidirect, or indirect forms of therapy can be utilized in treating hypernasality. It can be inferred that therapy for some subjects was aimed at the elimination of hypernasality. However, other speech disorders such as language problems and articulation disorders unrelated to hypernasality exist in some subjects and would have accounted for portions of the therapy services.

Surgery was included in the management for a majority of subjects. Retropharyngeal implants, combined palatoplasty/pharyngeal flap, and pharyngoplasty were the only surgical operations concerned directly with treatment of the velopharyngeal insufficiency. In some cases, T&A's were done in preparation for pharyngeal flap surgery to clear the pharyngeal area of limphoid tissue and were thus related to correction of velopharyngeal insufficiency. Other T&A's actually appeared to aggravate the problem and were performed for reasons other than the above. Other surgical operations performed were not concerned with the CPI problem.

The reported effect of management procedures on the speech characteristics of the subjects involves many variables. No direct correlation between any management technique and improvement in speech can be determined. However, evidence of the positive effect of both speech therapy and surgery, performed alone or in combination with each other, does exist. Seventy-five percent of those subjects having surgery directly related to decreasing velopharyngeal insufficiency have reported improvements in speech while 60% of those receiving speech therapy also report improvement.

It is difficult to attribute improved results in speech to any one technique since combinations of treatment are employed. These combinations are determined by the needs of individual subjects. Techniques often are used as adjuncts to each other. For example, speech therapy may be used to prepare an individual for surgery or to get optimal results from a surgical procedure already undertaken.

On the other hand, surgery may be performed to alleviate a condition that cannot be remediated behaviorally. Speech therapy may be resumed post-operatively although it is likely that, in some cases, recommendations for speech therapy may be traditional.

This discussion of treatment combinations relates to a very important aspect of CPI management, the sequencing of treatment as a function of time. A number of simplified chronological patterns may be identified. Speech therapy followed by an Oral Cleft Clinic recommendation of surgery is the most frequently occuring pattern. This same pattern occurs also with speech therapy performed post-operatively. One other major sequence is speech therapy followed by Oral Cleft Clinic examination and continued therapy for speech.

Inferences can be made from these patterns with reference to the type and severity of problems presented. Taking each one of the three treatment sequences, it is possible to conceive of hypothetical groups illustrating possible reasons for following the sequences. One group of children exhibit hypernasality of speech, receive speech therapy and show minimal improvement. Once examined at the Oral Cleft Clinic, it is determined that an anatomical/physiological deficit, not previously diagnosed, exists and should be dealt with surgically. Appropriate and successful surgery is carried out and the individual no longer experiences hypernasality. This hypernasality was probably physically based and as such, the problem was not responsive to speech therapy. Such a person may have gone through years of ineffective therapy. Another group may exhibit all of the above characteristics but after being treated surgically still evidence a need for speech therapy

services. The nature of this service is speculative. It is possible that surgical results were adequate but that hypernasality still exists because of the difficulty involved in adjusting to the new anatomical structure. In this case, speech therapy may help the patient adjust to the new structure. Or, surgical results may have been inadequate. Speech therapy may help reduce the remaining problem of hypernasality. Articulation or other problems unrelated to hypernasality may exist and warrant additional therapy post-operatively. There is also the possibility that post-operative therapy is traditionally expected and the person would thus be recommended to continue such services although they may no longer be of real value. The last hypothetical group has received speech therapy and has been recommended to the clinic because of questionable therapeutic results. A recommendation to continue speech therapy may imply one of several conditions. The subject may show no evidence of anatomical/physiological deficits and would therefore not be a candidate for surgical intervention. It may otherwise be discovered that no evidence of hypernasality exists but that other speech disorders such as denasality, articulation, or language disorders are present and require speech therapy.

The control that the patient (or parent) has over the management sequence is likely to have an effect on the resultant speech characteristics. Most subjects have no control over diagnosis of the condition and consequently accept any management procedure recommended. These procedures may not always be in the best interest of the patient. One subject related that the problem "involved mis-diagnosis of problem by school officials, unsuccessful speech therapy, and develop-

ment of poor self-image and defeatist attitude toward the problem. At age 20, subject was diagnosed as having a structural defect-shortened soft palate." On the other hand, subjects do have control over the acceptance or rejection of prescribed treatment recommendations and failure to comply with recommendations, especially surgical recommendations, will clearly alter results. In some cases, the subjects, or their parents, did not carry out surgical recommendations. One such subject began speech therapy in 1966 and was seen at the Oral Cleft Clinic in 1974 after eight years of therapy. Recommendations for surgery were made but, for unknown reasons, were not carried out. The subject is presently still receiving speech therapy.

The treatment sequences discussed are not necessarily complete. Since all subjects have been viewed retrospectively the data collection was conducted at different periods of each subject's treatment regime, there is no assurance that additional treatment will not occur. Any number of factors can alter the state of each subject's CPI condition at any point in time.

Implications for Further Research

From the research presented here, suggestions can be made concerning further study in the area of congenital palatal insufficiency. A projective study examining subjects systematically for the characteristics and variables identified would be of interest. In such a situation more control could be exerted over data collection and reliability and validity of information could be built in. A more in depth analysis of the specific speech and speech related

characteristics exhibited by CPI individuals as well as a delineation of techniques used in therapy would be helpful information. Perhaps comparative studies involving the use of different treatment procedures could be conducted. A longitudinal study could yield valuable information concerning the sequence and effect of management procedures as a function of time.

Many additional research possibilities exist in the area of congenital palatal insufficiency and the lack of currently available data suggest a real need for this research.

CHAPTER V

SUMMARY AND CONCLUSIONS

The purpose of this study was to analyze and describe characteristics of individuals with congenital palatal insufficiency and to determine information concerning problem onset, treatment, and referral. Twenty individuals seen at the Butterworth Hospital Oral Cleft Clinic of Butterworth Hospital, Grand Rapids, Michigan between September 1974 and June 1977 were selected as subjects. Questionnaires and medical files were used to obtain desired information. These information sources were analyzed and results were recorded.

Results indicate that the majority of subjects show indications of congenital palatal insufficiency during infancy and that speech therapists most frequently refer subjects to the clinic. All anatomical/physiological and related conditions studied are present in some members of the population. Velopharyngeal insufficiency with accompanying hypernasality of speech is the only condition which is evident in all population members.

Speech therapy is the most frequently employed management technique although its reported effect on speech is inconsistent. A number of surgical techniques for decreasing velopharyngeal insufficiency are used also in treating these subjects. Reported effects of these surgical procedures indicate high rates of speech improvement.

Various management sequences were reported. The majority of subjects follow one of three patterns of management. These patterns

involve (1) speech therapy followed by OCC examination and surgery (2) speech therapy followed by OCC examination, surgery, and continued speech therapy, or (3) speech therapy followed by OCC examination and recommendations for continued speech therapy.

Conclusions

The results of this study suggest that:

(1) Early identification of CPI is possible through a thorough examination of all anatomical/physiological and related characteristics presented.

(2) Velopharyngeal insufficiency and accompanying hypernasality of speech is the primary diagnostic feature of CPI.

(3) Speech therapists play an important role in the diagnosis and treatment of CPI individuals.

(4) Marked individual differences exist in the anatomical/physiological, management, and related characteristics in the CPI population.

APPENDIX A

COVER LETTER USED TO IDENTIFY STUDY POPULATION

WESTERN MICHIGAN UNIVERSITY

DEPARTMENT OF SPEECH PATHOLOGY AND AUDIOLOGY
Speech and Hearing Clinic
Phone 616 383-0963

KALAMAZOO, MICHIGAN
49008

August 1, 1977

Dear Parents:

We are currently doing research in the area of congenital palatal insufficiency. According to information available from the Oral Cleft Clinic files at Butterworth Hospital, your child fits the requirements of this study. We are seeking additional information concerning the surgical and speech therapy treatment of our subjects. If you would be willing to help us out in our research by answering a questionnaire, please let us know by mailing the enclosed postcard no later than August 19, 1977. A questionnaire will be sent to you within two weeks following our receipt of your postcard. All help will be greatly appreciated.

Sincerely,

Judith Dabkowski

Judith Dabkowski
Graduate student

Clyde R. Willis

Clyde R. Willis, Ph.D.
Professor

APPENDIX B

QUESTIONNAIRE AND EXPLANATORY LETTER

WESTERN MICHIGAN UNIVERSITY

DEPARTMENT OF SPEECH PATHOLOGY AND AUDIOLOGY
Speech and Hearing Clinic
Phone 616 383-0963

KALAMAZOO, MICHIGAN
49008

September 26, 1977

Dear Parents:

Enclosed is the questionnaire you agreed to complete to help us in our research on congenital palatal insufficiency. Information about specific subjects will remain confidential. No individual subject will be referred to by name in the results of the study. Infomation obtained from this questionnaire will be compiled and used to describe general characteristics of the population seen at the Butterworth Hospital Oral Cleft Clinic in the last three years.

Please be sure to fill in every answer. If you do not know the answer to any question, please indicate that the information is unknown or unavailable but DO NOT LEAVE ANY QUESTION UNANSWERED! Be sure to include all dates requested as these are very important to the results of this study. Please include approximate dates (especially the year) if the actual date is unknown. Feel free to use the back side of the questionnaire or additional paper if more space is needed to adequately answer any question.

Thank you for your time. Your help is greatly appreciated.

Sincerely,

Judith Dabkowski

Judith Dabkowski
Graduate Student

Clyde R. Willis

Clyde R. Willis, Ph.D.
Professor

P.S. PLEASE RETURN THE QUESTIONNAIRE IN THE ENCLOSED STAMPED ADDRESSED ENVELOPE NO LATER THAN OCTOBER17,1977.

IDENTIFYING INFORMATION

Name of subject_____Birthdate_____Phone_____
Parents' Name_____Address_____

HISTORY OF THE PROBLEM

When was the subject first examined at the Butterworth Hospital Oral Cleft Clinic?_____ How many times was the subject examined at the Oral Cleft Clinic after the first visit? Give month and year. _____ Who originally referred you to the Clinic? (check one) _____self or parent; _____Speech therapist; _____ Doctor (specify type); _____Family member; _____Other (specify profession). Describe as best you can the nature of the problem that resulted in referral to the Oral Cleft Clinic:_____

_____

_____

_____

_____

If there was special concern about the subject's speech, describe as best you can the nature of the speech problem. Include information on specific problems such as air leakage through the nose, too little air through the nose, difficulty with certain sounds, etc._____

_____

_____

_____

_____

_____

Describe any related problems. Include information on specific items such as drinking, sucking, swallowing, or blowing difficulties, breathing abnormalities, facial contortion, etc._____

_____

_____

_____

_____

When did the problem seem to originate? That is, was the problem always apparent? Or, was there a specific onset of the problem?_____

_____

_____

_____

_____

SURGERY

List all surgery the subject has had including the nature of the surgery, the date and place surgery was performed and the surgeon or surgery team who performed the surgery. List ALL surgery, regardless of its nature.

DATE	NATURE OF SURGERY	HOSPITAL	SURGEON

Did any of the above procedures have either a positive or negative effect on the subject's speech? That is, did speech improve or get worse after any of the above operations? If so, please identify the surgical procedure and describe the change in speech._____

SPEECH THERAPY

Identify the amount of speech therapy the subject has received by listing the year (or grade), setting (school, preschool, speech and hearing clinic, university clinic, private therapist, other), and the frequency (times per week).

YEAR OR GRADE IN SCHOOL	SETTING	FREQUENCY	GROUP OR INDIVIDUAL

Describe as best you can the effect speech therapy has had on the subject's speech_____

HEARING

Does the subject have a history of ear problems? _____yes _____no

If yes, describe the kinds of ear problems the subject has experienced

_____
_____
_____
_____

Identify below the treatment for ear problems the subject has had:

DATE	TREATMENT	SPECIALIST
_____	_____	_____
_____	_____	_____
_____	_____	_____
_____	_____	_____
_____	_____	_____

Describe the present status of the subject's ears and hearing. Add any other relevant information on the nature and course of the hearing problem.

_____
_____
_____
_____

ADDITIONAL INFORMATION

In the space provided below, add any other information that you feel may be relevant. You may want to include information on such items as: genetic counseling, use of prosthetic devices, school performance, social adjustment, self concept, etc._____

_____
_____
_____
_____

Form completed by_____
Relationship to subject_____

RETURN NO LATER THAN OCTOBER 17, 1977!

APPENDIX C

MASTER DATA CHART INCLUDING ANATOMICAL/PHYSIOLOGICAL, MANAGEMENT, AND RELATED CHARACTERISTICS FOR EACH SUBJECT

DATA CHART

	Anatomy						
Subj.	SMCP	Bifid uvula	Notch	Short palate	Deep vault	High Pal vault	Cleft lip
1	-	-	-	+	-	-	-
2	-	-	-	+	+	-	-
3	+	+	+	+	-	+	-
4	-	-	-	+	+	-	-
5	+	+	+	-	-	-	+
6	-	-	-	+	+	+	-
7	-	-	-	-	-	-	-
8	-	-	-	+	+	-	-
9	+	+	+	-	-	-	-
10	+	+	+	-	-	-	-
11	+	+	+	+	+	-	+
12	-	-	-	-	+	-	-
13	-	-	-	+	+	-	-
14	-	-	-	+	-	-	-
15	-	-	-	+	-	-	-
16	-	-	-	+	-	+	-
17	-	-	+	+	-	-	+
18	+	+	+	+	-	-	-
19	-	-	-	+	+	-	-
20	-	-	-	+	+	-	-

DATA CHART

	Physiology						
Subj.	VPI	Ear Infect.	Hearing Loss	Feed Diff	Swallow Diff	Suck Diff	Blow Diff
1	+	+	-	+	+	NI	NI
2	+	+	-	+	+	-	+
3	+	+	+	+	NI	-	+
4	+	-	-	-	-	-	-
5	+	+	-	-	-	-	-
6	+	-	-	+	+	+	+
7	+	+	+	+	+	+	+
8	+	-	-	-	+	-	-
9	+	+	-	-	-	-	-
10	+	-	-	NI	NI	NI	+
11	+	-	-	-	-	-	-
12	+	-	-	-	-	-	-
13	+	+	+	+	-	+	+
14	+	-	-	-	-	-	-
15	+	-	-	NI	NI	NI	NI
16	+	*NI	-	NI	NI	NI	NI
17	+	+	-	+	-	-	-
18	+	+	-	-	+	+	+
19	+	+	+	-	+	-	-
20	+	+	-	-	+	-	-

*No information = NI

DATA CHART

	Management									
Subj.	Spch ther	Pal plas	Phar flap	Phar plas	Retro ph imp	T&A	Lip rep	Ear surg	Push back	Prost
1	+	-	-	-	-	-	NA	+	-	-
2	+	-	-	-	-	+	NA	-	-	-
3	+	+	+	-	-	+	NA	+	-	-
4	+	-	-	-	+	+	+	-	-	-
5	+	-	-	-	-	-	NA	-	-	-
6	+	+	+	-	-	-	NA	-	-	-
7	+	+	+	+	-	+	NA	+	-	-
8	+	-	-	-	-	-	NA	-	-	-
9	-	-	-	-	-	+	NA	+	-	-
10	+	+	+	-	-	+	NA	-	-	-
11	+	+	+	-	-	+	+	-	-	-
12	+	-	-	-	-	-	NA	-	-	-
13	+	-	-	-	-	+	NA	+	-	-
14	+	-	-	-	-	+	NA	-	-	-
15	+	-	-	-	-	+	NA	-	-	-
16	NI	-	-	-	-	-	NA	-	-	-
17	+	-	-	-	-	-	+	+	-	-
18	+	-	-	-	-	-	NA	-	-	-
19	+	+	+	-	-	+	NA	-	-	-
20	+	-	-	-	+	+	NA	+	-	-

*Not applicable = NA

DATA CHART

	Related Conditions						
Subj.	Artic prob.	Lang. prob.	Decreas intell	Aller/ resp.	Mouth breath	Gen/ chrom	Congen abnorm
1	+	-	+	+	NI	NI	+
2	NI	-	-	+	+	+	-
3	+	+	+	-	+	+	+
4	+	+	+	-	NI	NI	-
5	-	-	-	-	NI	-	-
6	+	NI	-	-	NI	+	-
7	+	NI	-	+	NI	-	+
8	+	+	+	-	+	NI	+
9	NI	NI	-	-	NI	NI	-
10	NI	NI	-	NI	NI	-	-
11	-	-	-	+	NI	-	-
12	+	NI	-	+	NI	NI	-
13	+	NI	+	NI	NI	+	+
14	-	-	-	+	NI	-	-
15	-	-	-	+	+	+	-
16	+	+	+	NI	+	NI	+
17	+	+	+	NI	NI	-	-
18	+	+	+	NI	NI	NI	-
19	+	-	-	+	NI	-	-
20	+	-	-	-	NI	-	-

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