

The subject of oral clefts and their consequences has been dealt with technically. However, there has been little study of the untreated anomaly in adults, so that it is difficult to evaluate treatment programs. This study, developed to deal with the problem, offers some preliminary data and considerations.

UNOPERATED ORAL CLEFTS AT MATURATION

I. STUDY DESIGN AND GENERAL CONSIDERATIONS

Frank E. Law, D.D.S., M.P.H., F.A.P.H.A., and John T. Fulton, D.D.S., F.A.P.H.A.

CARL VON GRAEFE, often called the father of plastic surgery, published a lengthy dissertation about treatment of an oral cleft in 1820.¹ Eight years later, an Englishman named James Snell issued a book on oral cleft prosthesis which begins with a history dating back to 1579.² This review indicates that the early work was devoted to the repair of acquired clefts in adults, caused mostly by lues or gunshot wounds. Snell, who seems to be among the first to work with congenital clefts, presents some interesting case reports and technics involving both surgery and prosthesis for adults. Speech problems, with all their social and emotional overtones, are mentioned frequently as the compelling motive for treatment.

Snell laments the fact that many infants with oral clefts died of malnourishment, due to inability to suck. This fact, combined with the obvious speech problems of those who survived, probably accounts for the concentration on the surgical repairs of oral clefts in infancy, particularly when the development of anesthesia made better management possible.

The literature on surgery for oral

clefts has increased steadily in volume and quality from the early 1800's through to the present day.³ The development of this surgery is impressive in its imagination and skill but it has not solved the problems of oral clefts. For documentation, one has only to read the prosthetic literature devoted to rehabilitation of adults, which also has grown steadily since the time of Snell.⁴

A small, but growing literature exists concerning the social and emotional effects of facial deformity. At the same time, speech pathologists are concerned with the social and emotional consequences of speech disorders. Thus the individual who is born with a cleft palate and lip suffers a twofold problem. Present indications lead to the conclusion that afflictions of this nature result in a variety of consequences which bear heavily on the adjustment of the individual. However, it seems to be agreed that ease or difficulty of maintaining personal equilibrium is based primarily on the personality of the individual involved, and that certain individuals seem much better able to bear up under such a deformity than do others.

Good speech and appearance are the

compelling motives for treatment and the coalescence of this purpose in the last 20 years has led to the multidiscipline or so-called "team" approach.⁵ Treatment continues to concentrate on young children and successful surgery still is the goal, but the biosocial nature of the oral cleft now is recognized and a battery of professional skills help in the diagnosis and treatment plan.

The voluminous literature on oral clefts is devoted essentially to the techniques of treatment. Studies of the anomaly itself are few in number. What may be called the "life history" of the untreated oral cleft remains obscure. Its "birth" has not been defined precisely⁶; its growth and development are being studied longitudinally but, so far, have been reported only through early childhood.⁷ Finally, there is no systematic description of the final phase of the life history—the untreated anomaly in adults.

This lack of knowledge concerning the latter aspect of the life history makes it difficult to evaluate treatment of these defects. Treatment for an oral cleft, in a biosocial context, can be evaluated adequately only in the mature individual. Rehabilitation includes the reinstatement of the individual into his environment, equipped to function normally in this environment to the limit of his capacity. A proper yardstick seems basic to the answering of certain questions of importance to this kind of evaluation.

1. What is the normal outcome of an oral cleft if allowed to mature without treatment?

2. How does an adult with an untreated anomaly differ from his peers in physical function, social adjustment, and emotional health?

3. How does such an adult compare with one who has received conventional treatment?

These important questions are the basis of the present study

Methods and Materials

A major problem in establishing this study was the location of a suitable

group of cases. The primary criteria for inclusion in the study group were that the person (1) must have reached maturation, 15 years of age or over, and (2) have a palatal cleft which had never received surgical or prosthetic treatment.

The interest and cooperation of the Insular Department of Health made it possible to locate a sizable group of such cases on the Island of Puerto Rico and the study was organized, finally, around this group.

A study team of five dentists, a speech pathologist* and an orthodontist† was assembled with adequate supplies and equipment. This team visited Puerto Rico in June, 1957, and June, 1958. Upon arrival in Puerto Rico this unit was augmented with dentists, social workers and other personnel from the Insular Department of Health and local health centers. The assistance of this personnel was invaluable in organization, interviewing, interpretation, patient management, and the clinical aspects of the study.

Case finding was accomplished, prior to the team's arrival, by the Bureaus of Crippled Children and Dental Health of the Puerto Rico Department of Health assisted by the local Office of Vocational Rehabilitation.

The immediate objectives of the study were:

1. To determine the facial and nasopharyngeal growth and development of these untreated cases.

2. To evaluate their speech and voice quality and, in this area, how well they had adapted to the handicap.

3. To compare the hearing ability of such persons with normal or average groups of the same ages.

4. To study their medical history as it may relate to the palatal condition.

5. To establish the social status of these palatal cleft cases within their group and

* Dr. McKenzie W. Buck, head, Speech and Hearing Clinic, University of Florida, Gainesville, Fla.

† Dr. J. Daniel Subtelny, head, Department of Orthodontics, Eastman Dental Dispensary, Rochester, N. Y.

some of the social and emotional problems encountered by these individuals.

To achieve these objectives the following procedures were applied to each study case.

Cephalometric radiographs in various positions and when pronouncing certain vowels were taken. Recording of speech was taped and hearing was tested with a pure tone audiometer. A dental examination and impressions of the upper and lower arches including the palatal defect were made. Intraoral and extraoral color photographs were taken. Interviews were conducted in Spanish by local social workers. The social interview form was developed by a social psychologist* and pretested in Puerto Rico before the actual study began. Medical examinations were conducted on the first study group but, as the results were completely negative, they were not included for the second group. Procedures were carried out at five different health centers in order to obtain complete coverage of the Island.

Data have been collected on 124 native Puerto Ricans during the two study periods. These individuals range from 15 to 57 years of age and are almost equally divided between male and female. Untreated clefts of the palate were present in 51 of these people; eight others had clefts involving only the lip and alveolar process; 29 had palatal clefts with prior surgical repair, and were included for comparison of treated and untreated cases. The other 36 people examined had normal palates and were included in the study as controls for normal growth and speech of Puerto Ricans. They were selected at random from available adults at the examination centers.

Table 1 gives the distribution by age and sex of the 88 treated and untreated

cases of oral clefts. It is interesting to note that 90 per cent of this group were between the ages of 15 through 34 years. Some of the factors which may account for the paucity of patients above age 34 are: these patients may have been working and were thus unable or unwilling to attend the clinic; because of their age, they may have felt that treatment was unlikely or they were uninterested in treatment; they may not have survived to these ages, or, because of the oral defect, they may have isolated themselves.

It is of further interest that, in this group, twice as many females had posterior clefts as males, while four of the five bilateral clefts occurred in males. Complete unilateral clefts and clefts involving only the lip and alveolar process were fairly equally divided between the sexes.

Table 2 shows the distribution of cases by type of cleft.

In the eight cases in which the oral cleft involved only the lip and the alveolar process there were none where surgical repair had been attempted on the defect of the alveolar process. The lip had been treated surgically in five instances. This study is interested pri-

Table 1—Distribution by Age and Sex of 88 Patients with Oral Clefts—Puerto Rico, 1957 and 1958

Age Groups	Male	Female	Total
15-19	23	20	43
20-24	9	10	19
25-29	3	7	10
30-34	4	3	7
35-39	-	1	1
40-44	2	1	3
45-49	2	-	2
50-54	-	-	-
55-59	3	-	3
Total	46	42	88

* Dr. S. Stephen Kegeles, research social psychologist, Division of Dental Public Health, Public Health Service, Department of Health, Education, and Welfare, Washington, D. C.

Table 2—Distribution by Type of Cleft of 88 Cases of Oral Clefts—Puerto Rico, 1957 and 1958

Posterior clefts involving hard and soft palates	33
Unilateral clefts involving lip, alveolar ridge, hard palate, and soft palate (right 10—left 32)	42
Bilateral clefts involving lip, alveolar ridge, hard palate, and soft palate	5
Unilateral clefts involving lip and alveolar ridge (right 2—left 6)	8
Total	88

marily in clefts involving the palate so these eight cases were dropped from further consideration.

It was decided to include, in the untreated palate group, those cases in which the lip had been closed surgically but the palate had never received surgical or prosthetic treatment. Table 3 lists the 47 cases of clefts involving both lip and palate. It shows that 40 of the 47 cases (85 per cent) had received lip repairs. Table 4, presenting the data on palate repairs, shows that only 18 (38 per cent) of the 47 cases had repairs attempted on the palate. Of the 33 cases of posterior clefts, without lip involvement, shown in Table 4, 22 (64 per cent) had no surgical treatment.

General Findings

These unoperated cleft palate cases will be discussed briefly in four general areas: (1) physical functioning, (2) facial growth and development, (3) communication, and (4) social and emotional consequences:

(1) Physical Function—The cases available for study had lived to maturation so they obviously had survived the problems of breathing, sucking (eating) and swallowing that are associated with

this defect. Other physical functioning appeared quite normal. No information is available on the prevalence of cleft palate in Puerto Rico or on the differential mortality of such conditions, and it cannot even be surmised how many failed to survive. It was the clinical impression of the team that the individuals studied were of average health and physical development when compared with the control group and others in the area. The medical examinations on the 1957 group were essentially negative and, as mentioned earlier, were not repeated on the second group. On questioning these patients seemed to have no more upper respiratory infections than the normal group. The turbinates were usually abnormally enlarged and of a purplish hue, but the eustachian tubes were usually patent and apparently functioning normally.

As far as could be determined clinically these untreated cleft palate cases did not appear to suffer the ailments commonly associated with surgically repaired cases. The health and physical functioning of the untreated group seemed equal to that of individuals with normal palates.

(2) Growth of Face—The facial growth and development of the unoperated cleft palate cases appeared to have progressed on a normal pattern and

Table 3—Distribution by Lip Repair of 47 Cases of Unilateral and Bilateral Oral Clefts—Puerto Rico, 1957 and 1958

	Lip Unoperated	Lip Operated	Total
Unilateral			
Right	2	8	10
Left	4	28	32
Bilateral	1	4	5
Total	7	40	47

achieved near normal skeletal dimensions. The study team saw no external evidence of abnormal facial growth. The dental arches appeared normal, except for the fault itself, and in harmony with the facial pattern.

Radiographically the vertical growth, the anteroposterior growth and the lateral growth of the maxilla or upper jaw, as well as mandibular length and position, were similar in adults with non-operated cleft palates and normal adults. They did not exhibit the deficiencies of the maxilla sometimes seen, at this age, in cases with an apparent growth interference of the upper jaw.

Visually, there did not appear to be any growth disturbance in the upper part of the face in these patients. Generally, there was no major discrepancy between the upper and lower parts of the face. In the few cases showing some malrelation it was thought to be the genetic pattern of the individual.

The cases of complete clefts involving the lip, alveolar process and palate appeared to exhibit a more normal maxillary dental arch contour when the lip was operated on at a later age than do the cases that have been operated on in infancy.

The tissues of the velopharyngeal area,

in these untreated adults, appeared to be much less than normal and gave the impression of some atrophy. This may be atrophy related to dysfunction or malfunction or there may be lack of soft palate tissue at birth. The majority of these appeared to have an insufficiency of soft palate tissue and an inadequacy of soft palate mobility.

(3) Communication—Clinical impressions and a tentative analysis of speech data confirm the statement regarding the similarity in maxillary and mandibular growth between nonoperated cleft palates and normal adults.

Studies in the United States⁸ indicate that the anteroposterior dimensions of the oral cavity are shorter in repaired cleft palate groups than in normal subjects. This anteroposterior dimension did not differ significantly from the normal in the unoperated Puerto Rican group.

Tongue length is determined by data for the tongue area during vowel phonation and at rest position. The tongue measurements of the unrepaired cleft palate group appeared to fall within normal structure measurements.

Repaired cleft palate subjects appear to employ the same tongue positioning during phonation as do normal cases. There is some evidence in the unrepaired group in Puerto Rico that the tongue tends to be carried in a much higher position in the oral-nasal cavity. Lack of restrictive structures superiorly to guide the tongue may explain this positioning. Many of the sounds were being made in the unrepaired complete cleft with tongue contact on the superior border of the nasal area.

In repaired cleft palate groups the movement of the posterior pharyngeal wall during vowel phonation is more restricted than in normal groups. In the unrepaired cases in this study, measurements, thus far, indicate average to better than average pharyngeal wall movement for all vowels.

Lip and mouth opening during phona-

Table 4—Distribution by Repair of Palate of 80 Cases of Unilateral, Bilateral and Posterior Oral Clefts—Puerto Rico, 1957 and 1958

	Palate Unoperated	Palate Operated	Total
Unilateral clefts			
Right	4	6	10
Left	23	9	32
Bilateral clefts	2	3	5
Posterior clefts	22	11	33
Total	51	29	80

tion among United States repaired cleft palate subjects, as compared to normal United States subjects,⁸ reveals no statistically significant difference. It is somewhat difficult to measure lip opening in unrepaired cases, because of the lack of movement of the superior structures. However, there is an indication, thus far, that the mouth opening (measurement of distance between the mandible and the maxilla) in the unrepaired complete cleft palate group seems to be approaching a wider opening.

Clinically, it was observed that individuals having rather large clefts of the palate frequently exhibited good speech and voice quality. There was much less nasal quality than one would expect when account is taken of the extent of palatal abnormality. The more restricted the cleft, the more positive the judgments of nasality. The individuals who had a complete unrepaired cleft from the lip through the soft palate had ratings of near normal voice qualities; i.e., little if any nasality was observed. This lack of nasality might be explained by the fact that complete clefts with no repair had but one resonating cavity. Individuals who had an unrepaired cleft of the hard and soft palates tended to be judged more nasal, and individuals with a cleft of the soft palate only had a high consistent rating of nasality. The more separation of the oral and nasal cavities, in these cases, the more well defined were the auditory symptoms of nasal quality.

Six well trained speech and hearing personnel have listened to the tape recordings obtained on the Puerto Rican subjects. Quite frequently it is necessary to play the tapes of American patients backward to avoid confusing nasal emission with poor articulation. In view of the fact that the Puerto Rican tapes were in Spanish (which none of the listeners understood) there could have been no adverse effect upon the reliability of judgment. These speech pathologists were

listening only to the production of vowel and consonant sounds and not to the correct production of a complete word.

In many instances, the individuals who had complete and wide clefts produced all of the speech sounds except (k) and (g). The production of the (k) and (g) sounds requires the cooperation of the soft palate tissues to produce the plosion in the posterior portion of the mouth. The omission of (k) and (g) sounds for all types of clefts was consistent. The production of speech, however, by clefts of the hard and soft palate posterior to the premaxilla, seemed to be superior to the speech produced by many patients who have had a complete closure of the cleft. It is quite possible that these Puerto Rican patients, who had the opportunity of experiencing more normal facial growth, enjoyed the advantage of good occlusion between the maxilla and the mandible, thus assisting them in the production of the speech sounds.

Although the environment of the hearing tests was not ideal the results may be considered clinically accurate.

Two-thirds of the unoperated cleft palate patients receiving audiometer tests had essentially normal hearing acuity. The one-third registering hearing difficulties was divided equally between monaural and binaural hearing losses. The degree of hearing loss in the monaural group was recorded as mild, 66 per cent; moderate, 22 per cent; severe, 6 per cent; and deaf, 6 per cent. In the binaural group the results were 35, 56, and 11 per cent, respectively, with no patients recorded as deaf.

The monaural hearing loss recorded would have little effect upon ability to learn and reproduce speech sounds. A number of the cases tested were approaching an age level of natural auditory nerve deterioration which tends to increase the per cent of cases with binaural hearing loss.

There were 12 cases needing special assistance with their hearing in regard

to lipreading and speech production. This is a considerably higher proportion than that found among normally-structured individuals. However, when one considers the number of hearing losses obtained on repaired cleft palate cases, this ratio for unrepaired cleft palates is very small.

One might speculate from this preliminary investigation that the postponement of surgical repair of the soft palate tissues appears to be of considerable assistance in maintaining adequate social hearing.

(4) Social Problems—The small exploratory study in Puerto Rico was directed toward gathering from the cleft lip and palate individuals their own perceptions of the consequences during their lifetime of having been so afflicted.

An attempt was made to arrive at data which would help toward an understanding of the types of situations which the afflicted individual faced during his life from infancy to adulthood. Five periods of potential stress were chosen concerning which the individuals were asked about their perceptions. These were: (1) the recollection of early home life, including the present memory of treatment by parents as compared to that given siblings in the home; (2) school life, including difficulties found in relation to teachers and schoolmates; (3) early social life, including desire for, and participation in, children's social activities; (4) job history, including feelings about jobs, and the ease or difficulty of securing and holding jobs; and (5) present social life, including family relations, as well as desire for and participation in community social life.

The following types of analyses will eventually be made: (1) a comparison of treated, untreated, and normal control cases according to the five dimensions noted above; (2) a comparison of the five dimensions for differences between individuals with greater or lesser facial deformity, and greater or lesser

speech disturbance after such criteria have been demarcated; and (3) a suggestion of further studies which lead from these frankly exploratory data.

It has been difficult to arrive at anything more than impressions of the data in the time available. However, it looks very much as though the individual born with a cleft palate encounters situations with great social and emotional consequences almost from the beginning, and which continue almost unabated throughout his life.

The cleft palate cases recalled, in adulthood, that they were treated quite differently than their siblings in the home, that they were left alone, rarely punished, and received, in many instances, greater amounts of love than their brothers and sisters. There were very few cases of noted parental rejection. In most cases, these individuals reported being the favored child.

When the child went to school, he seemed to find that again he was treated quite differently from the other children. Thus, he reports that he had far more difficulty in learning the necessary subject matter, especially in learning English, and in reading out loud. He was quite frequently the butt of ridicule by other children in and out of the classroom, and the teachers attempted to protect him from this ridicule. In certain instances subjects reported feeling quite hurt by what they saw as overprotection.

Discussion

It must be stressed that the general findings presented in this paper are based on clinical observations, impressions of the examining team, and some preliminary study of the data. In spite of these limitations several tentative conclusions seem warranted.

These nonoperated cleft palate cases appear to have developed well and to have followed normal facial growth patterns.

The maxillary growth and development in these unrepaired cases appeared to approach that of normal adults more closely than many of the repaired cases. Clinically, the growth of the upper part of the face and of the dental arch was closer to normal in the nonoperated cases than in cases with early surgical treatment of the palate.

Measurements of the unoperated cases indicate better movement of the posterior pharyngeal wall during vowel phonation than was found in repaired cases.

These untreated patients reported no more respiratory illnesses than the control groups and although the turbinates were consistently abnormally enlarged the eustachian tubes were usually patent and apparently functioning normally.

Individuals in the study group had adapted rather well to speech difficulties. All types of palatal clefts consistently omitted (k) and (g) sounds. Unrepaired cases of complete clefts from the lip through the soft palate had near normal voice quality while those with a cleft of the soft palate only had the most nasality. The speech sounds produced by patients with untreated clefts involving the hard and soft palates appeared superior to the speech sounds of patients with surgical closure of the cleft.

Although a higher proportion of the study cases than normal required special assistance with hearing problems, this ratio was lower than that found among repaired cases.

Social and emotional problems, resulting from a speech disorder, a facial deformity, or often from both, develop early in life and continue to plague the

individual. However, these people reported that as children at home they were favored over their siblings and at school the teachers tended to protect them and in many instances to be over-protective. The great majority had considerable difficulty in school.

It is estimated that about 250 adults with oral clefts should be available on the Island for study. It is planned to continue the project to locate and collect data on a sufficient number of cases to produce a truly representative sample of this group.

Additional areas for productive investigation, in these groups, are becoming apparent as the data on hand are studied. These areas include: the genetic aspects of oral clefts, results of treatment at different ages and by various methods, speech training problems and a variety of emotional factors.

REFERENCES

1. Graefe, C. F. von. Die Gaumennath, ein neu entdecktes Mittel gegen angeborene Fehler der Sprache. *J. Chir. u. Augenh.*, Berlin 1:1-54, 1820.
2. Snell, James. Observations on the History, Use and Construction of Obturateurs. London, England: Callow and Wilson, 1828, 106 pp.
3. Cannon, Bradford, and Fisler, David. Plastic Surgery: Harelip and Cleft Palate. *New England J. Med.* 245:179-185 (Aug.), 1951.
4. Onachilla, M. M. The History of the Non-Surgical Treatment for Acquired Palatal Deformities. Unpublished Master's Thesis. Pennsylvania State College, 1946.
5. Baker, H. K. The Multidiscipline Approach to the Treatment of the Child with Cleft Palate. *J. Internat. Coll. Surgeons* 24:367-369 (Sept.), 1955.
6. Fulton, J. T. Closure of the Human Palate in Embryo. *Am. J. Obst. and Gynec.* 74:179-182 (July), 1957.
7. Pruzansky, Samuel. Description, Classification, and Analysis of Unoperated Clefts of the Lip and Palate. *Am. J. Orthodontics* 39:590-611 (Aug.), 1953.
8. Buck, McKenzie W. Facial Skeletal Measurements and Tongue Carriage in Subjects with Repaired Cleft Palates. *J. Speech & Hearing Disorders* 18:121-132 (June), 1953.

Dr. Law is chief, Operational Research Branch, Division of Dental Public Health, Public Health Service, Washington, D. C., and Dr. Fulton is professor of dental epidemiology, University of North Carolina, Chapel Hill, N. C.

This paper was presented before a Joint Session of the Dental Health and Maternal and Child Health Sections of the American Public Health Association at the Eighty-Sixth Annual Meeting in St. Louis, Mo., October 28, 1958.