

Longitudinal Study of Skeletal and Soft Tissue Profile in Children with Unilateral Cleft Lip and Cleft Palate

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Previous growth studies on the facial profile of cleft lip and palate populations have been limited to the hard tissues. Yet it is clearly apparent that distortions, deficiencies, or overgrowth in the integumental profile are additional and possibly independent variables that contribute to the total cosmetic and functional results of facial reconstruction. For these reasons the present study was designed to evaluate the characteristics of the integumental profile from a longitudinal series of roentgenocephalometric films on a population of patients with complete unilateral clefts of the lip and palate.

Specifically, information was sought to answer a series of questions that Subtelny (9) had previously posed in studying the same problem in a normal population: a) What are the longitudinal characteristics of the soft tissue profile? b) Are there differentials in growth associated with different aspects of the soft tissue profile? c) If there are differentials in soft tissue growth, how do these affect the configuration of the total profile? d) Is the soft tissue profile closely related to the underlying skeletal profile?

Methods and Materials

The roentgenocephalometric films utilized in this report were selected from the serial growth study in continuing progress since 1949 at the Cleft Palate Center of the University of Illinois. The methodology has been previously described by Pruzansky and Lis (6).

The sample included 21 subjects with complete unilateral cleft lip and palate for whom serial cephalometric films were available from six months to seven years of age. The population included 14 males and seven females. In 11 subjects the cleft was on the left side of the face and on the right in the other ten. The accumulated data were compared with the normative data on the soft tissue profile published by Subtelny (9).

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It should be noted that Subtelny described his samples obtained from the Bolton Study at Western Reserve as "lateral cephalometric roentgenograms taken with the teeth in occlusion." Our films were obtained with the mandible at "rest position" during the first three years of life, since occlusion could not be obtained in a reliable fashion during those earlier ages. After about three years of age, the lateral films were in occlusion.

The tracing included the outline of the soft tissue structures forming the profile as well as the underlying osseous structures. The landmarks identifying the points on the soft tissues and facial skeleton were similar to those described by Subtelny (Figures 1 and 2). Standard methods of

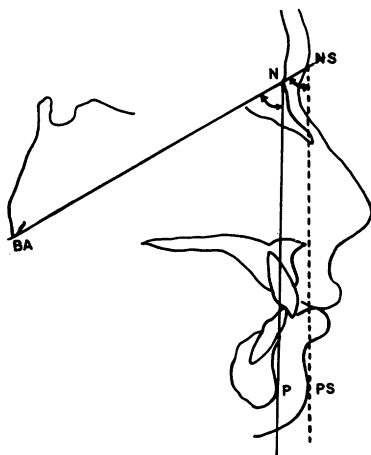


FIGURE 1. Angular method of determining the relative anteroposterior position of the skeletal chin (angle Ba—N—P) and the integumental chin (angle Ba—Ns—Ps) in reference to the cranial base line. (Courtesy of Dr. J. Daniel Subtelny)

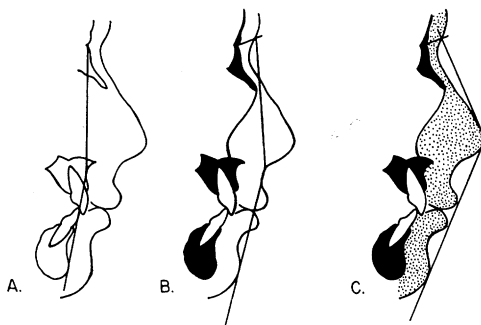


FIGURE 2. Methods of evaluated facial convexity. A, Convexity of the skeletal profile (nasion—point A—pogonion). B, Soft tissue profile convexity (soft tissue nasion—subnasale—soft tissue pogonion). C, Total soft tissue profile convexity, including the nose (soft tissue nasion—tip of nose—soft tissue pogonion). (Courtesy of Dr. J. Daniel Subtelny)

superimposition and measurement were employed to evaluate changes on a time basis. Means and standard errors were obtained for the cleft palate population at specific age levels. The data were then compared with the findings available for the normal.

Findings

To facilitate comparisons between the data in this study and the previously cited report on a normal population (9), the sequence of presentation will be similar to that employed in that publication.

MANDIBULAR PROGNATHISM. Protrusion of the chin point was measured by angular relation to the cranial base. The cranial base line was constructed by connecting basion to nasion. The chin-point, pogonion, was connected to nasion and the angle basion-nasion-pogonion was formed by the intersection of the two lines. To satisfy the need for corresponding landmarks for analysis of the soft tissue profile, the cranial base line was extended anteriorly to intersect overlying soft tissue and thus establish *soft tissue nasion* (NS). The most anterior point of the soft tissue chin was designated as *soft tissue pogonion* (PS). The prognathism of the soft tissue profile was evaluated on the basis of the opening or closing of the angle formed by basion-soft tissue nasion and soft tissue pogonion (Figure 1).

Changes in the prognathism of the skeletal chin as a function of growth. The skeletal chin tended to become more prognathic with increments in age. The increase in prognathism was observed after the second year and continued through the fifth year of life. In the interim between the fifth and seventh year, a decrease in prognathism was recorded. An equivalent number of subjects was not available for the later years as in the earlier age group.

As shown in Table 1, the tendency toward increased mandibular prognathism, though quite apparent, was notably less for the cleft palate group (mean: 2.1°) when compared to the normal male group (mean: 4.6°). Comparison with the normal female group demonstrated even greater disparity, since at every age level studied the normal female was more prognathic than the normal male. Although the pattern of change

TABLE 1. Mean angular measurements of mandibular position.

AGE IN YEARS	SKELETAL PROGNATHISM			SOFT TISSUE PROGNATHISM		
	CLEFT PALATE	NORMAL		CLEFT PALATE	NORMAL	
	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE
½	51.7	54.0	57.0	53.9	55.5	57.5
1	51.9	56.2	57.3	53.9	57.5	57.7
2	52.2	57.6	59.7	53.6	58.1	59.4
3	52.7	58.4	59.7	53.5	59.1	60.6
4	53.7	58.2	60.4	55.1	60.0	61.9
5	54.5	58.2	60.4	55.5	60.3	61.8
6	52.5	58.0	60.5	55.5	60.1	61.9
7	53.8	58.6	60.1	56.6	60.0	62.0

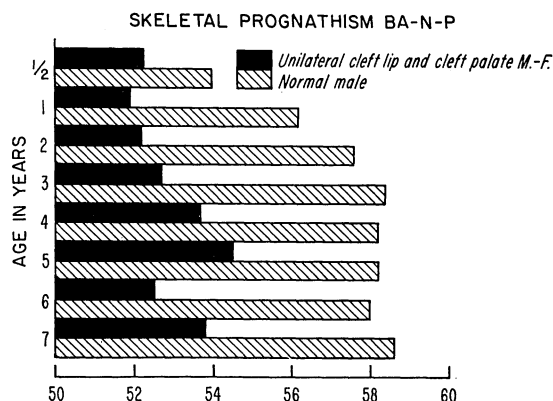


FIGURE 3. Amount of skeletal prognathism (in degrees) for cleft and normal subjects according to age.

was somewhat similar to that observed in the normal sample, the cleft lip and palate group tended to be less prognathic from birth through seven years of age when compared to the normal sample during analogous periods. The average measurement of mandibular prognathism at five years of age for the cleft lip and palate group was comparable to that of the normal group at six months of age (Figure 3).

Changes in the prognathism of the integumental chin as a function of growth. The integumental chin also tends to become more prognathic with an increase in age, particularly after the age of four.

When the cleft palate group was compared to the normal group, it was quite obvious that the integumental chin was less prognathic at every age level studied (Figure 4). Smaller increments in mandibular prognathism were noted for the cleft palate group (mean: 2.7°) from six months through seven years of age when compared to the normal (mean: 4.5°).

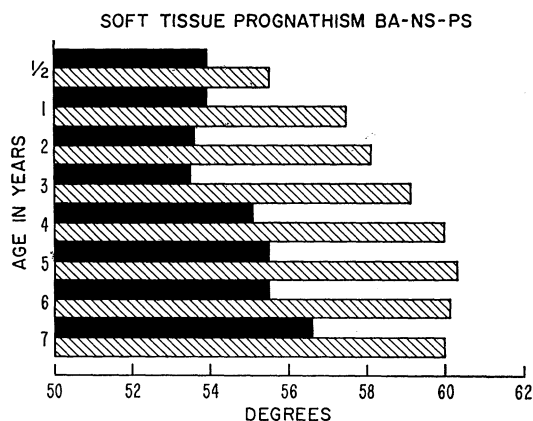


FIGURE 4. Amount of soft tissue prognathism (in degrees) for cleft and normal subjects according to age.

To restate the findings in another way, the average measurement for the anterior location of the integumental chin at five years of age in the cleft palate group was similar to that in the normal group at six months of age.

Comparisons between skeletal and soft tissue measurements of mandibular prognathism as a function of growth. Mean angular measurements of mandibular position for skeletal and soft tissue prognathism indicated that the chin became more prominent with advancing age and tended to assume a more forward position with relationship to the cranial base. The same pattern of change was noted in both skeletal and soft tissue profile measurements (Table 1). When compared with normative data, the cleft lip population revealed a skeletal and integumental chin that was more retrusive at every age level studied. The degree of increase in mandibular prognathism was found to be less for the cleft palate group during analogous periods of comparison. Thus it is apparent that the cleft palate skeletal and integumental chin, compared to the normal group, was found to be less protrusive at six months and persisted in this manner throughout the periods studied (Figures 3 and 4). This finding is statistically significant at the .1% level.

CONVEXITY OF THE SKELETAL PROFILE. The angle of convexity (β) (nasion-point A-pogonion) was employed to evaluate the relative antero-posterior position of the upper face to the cranium and lower face. The smaller the angle measured, the greater the convexity of the facial profile (Figure 2A).

Average angular measurements revealed a rapid decrease in the convexity of the skeletal profile from six months to three years of age. From three to five years of age a continued reduction in skeletal profile convexity is noted. Thus during the first five years of life there is notable straightening of the cleft palate skeletal profile. Between the fifth and seventh years, some slight increase in the skeletal profile convexity is observed (Table 2).

At every age level measured, skeletal facial convexity was significantly greater for the cleft palate population than for the normal. In addition,

TABLE 2. Average measurements of the degree of convexity of the facial profile.

AGE IN YEARS	SKELETAL PROFILE CONVEXITY (N-A-P)			SOFT TISSUE CONVEXITY (NS-SN-PS)			TOTAL PROFILE CONVEXITY (NS-NO-PS)		
	CLEFT PALATE		NORMAL	CLEFT PALATE		NORMAL	CLEFT PALATE		NORMAL
	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE
½	152.2	160.0	160.0	160.3	161.0	161.2	140.4	138.0	141.2
1	156.5	164.2	164.0	163.2	161.6	163.7	145.6	139.5	142.4
2	159.4	167.5	168.9	163.6	163.9	165.1	146.3	142.5	142.3
3	160.3	170.0	169.0	165.5	165.0	165.3	146.3	142.0	140.9
4	162.1	170.3	169.3	166.7	163.0	162.9	147.6	140.7	139.1
5	163.9	170.6	171.6	168.2	162.1	164.7	147.3	138.5	139.6
6	162.0	171.6	170.4	168.0	162.2	161.0	144.8	138.7	136.8
7	161.5	172.9	171.5	165.3	161.6	161.1	142.5	140.3	135.7

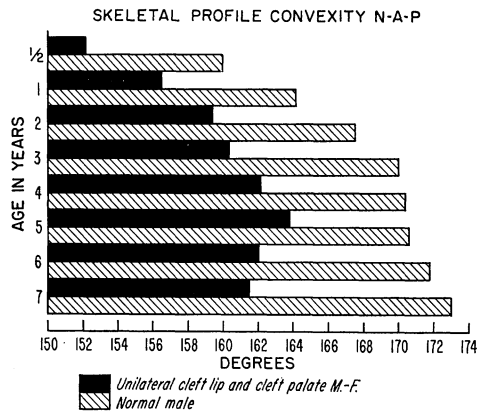


FIGURE 5. Amount of skeletal profile convexity (in degrees) for cleft and normal subjects according to age.

the growth changes observed indicated that the facial convexity tended to diminish to a greater extent in the normal than it did for the cleft palate sample (Figure 5).

To determine what role point A played in reducing the angle of convexity, the angle SNA was measured for each cleft palate subject. This angle remained constant during the periods analyzed. Thus the discernible decrease in the skeletal profile convexity was a function of the greater forward growth of the mandible in relation to nasion and point A. Comparative longitudinal measurements for the normal were not available.

CONVEXITY OF SOFT TISSUE PROFILE. Two methods were employed to evaluate the changes in the convexity of the soft tissue profile. One method did not include the nose and the other method did. The first was an assessment of the profile formed by the soft tissue areas that closely paralleled the skeletal cephalometric landmarks employed in measuring convexity of the skeletal profile.

The second method employed the tip of the nose as the central point of the angle, making this soft tissue landmark the frame of reference in estimating convexity. The marked influence of the nose in the soft tissue profile is readily recognized and in the cleft palate subjects its role in this area of evaluation becomes even more apparent (Figure 2C).

Soft tissue profile convexity excluding nose (NS-SN-PS). Average measurements of the convexity of the soft tissue profile, excluding the nose, were found to show a reduction in the profile convexity. Although the pattern of change for both groups appears to be similar, some differences were noted. Both groups demonstrated reduction in the soft tissue profile convexity up to three years of age. The cleft palate group, after three years, exhibited a continued reduction in the soft tissue profile convexity through the sixth year. Between the sixth and seventh year, a

notable increase was observed. By comparison, the normal group demonstrated an apparent increase in profile convexity, from the third through the seventh year (Table 2). Thus, the cleft palate group tends to reflect a soft tissue profile that was less convex, at most ages, than that recorded for the normal group (Figure 6). This finding was statistically significant at the 5% level.

In the normal the following was noted. Longitudinal changes for soft tissue landmarks in the upper face were not analogous to those exhibited by underlying skeletal points. The skeletal profile became less convex from six months through seven years while the soft tissue profile became less convex up to three years of age, after which it exhibited a notable increase in the soft tissue profile convexity until seven years of age. The cleft palate population, on the other hand, reflected a reduction for both soft tissue and skeletal profile from six months through five years of age. Thereafter until seven years a slight increase in convexity was noted for both.

The difference noted in the behavior of the soft tissue profile convexity between cleft palate and the normal groups indicates that an independent variable is actively affecting the contours of the upper lip during the early years of growth. For the cleft palate group, the soft tissue profile, excluding the nose, tends to become straighter (Figure 7).

Soft tissue measurements of total profile convexity including the nose (SN-PO-PS). The soft tissue profile, including the nose, was significantly less convex for the cleft palate sample than for the normal population during the age period from six months to seven years (See Table 2 and Figures 8 and 9).

The deviant shape of the nose in patients with cleft of the lip accounted for the variance of the soft tissue profile of this population and the difference from the normal comes as no surprise (Table 3).

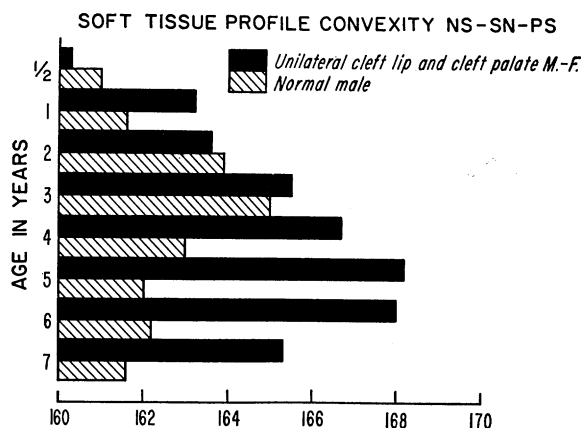


FIGURE 6. Amount of soft tissue profile convexity (in degrees) for cleft and normal subjects according to age.

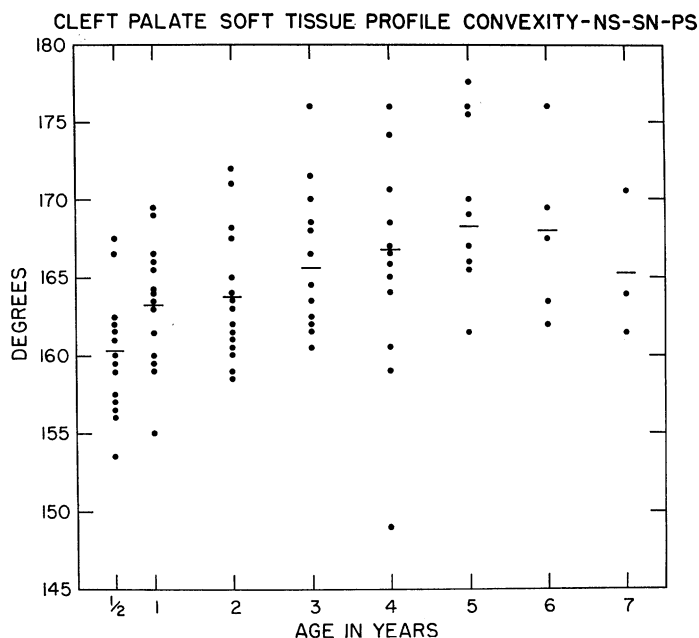


FIGURE 7. Individual variation (dots) and mean values (—) for degrees of soft tissue profile convexity for the cleft palate subjects according to age.

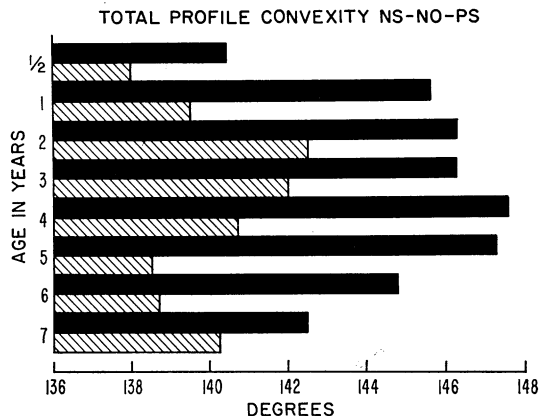


FIGURE 8. Amount of total profile convexity (in degrees) for cleft and normal subjects according to age.

As previously noted, the soft tissue profile, excluding the nose, tended to become less convex with age in the cleft sample. This observation was all the more striking in view of the reduced mandibular prognathism in the cleft subjects. Apparently, then, the soft tissue in the subnasale area behaved independently of its skeletal counterpart, point A.

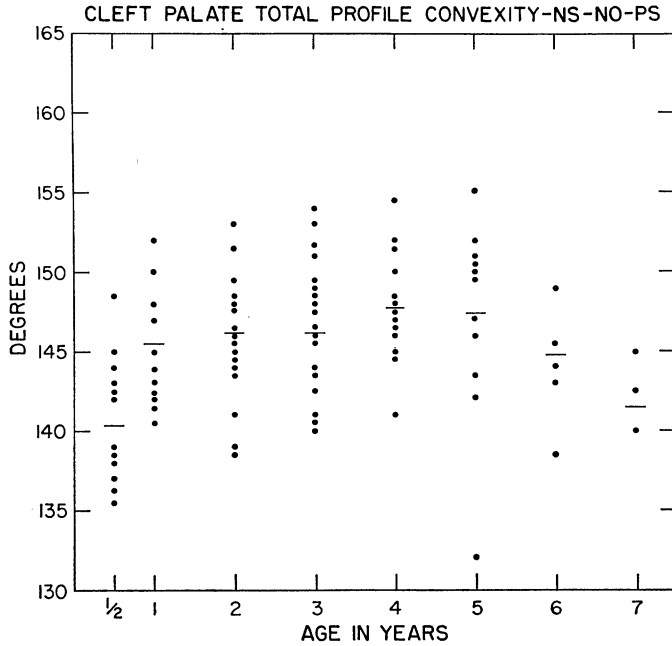


FIGURE 9. Individual variation (dots) and mean values (—) for degrees of total profile convexity for cleft palate subjects according to age.

TABLE 3. Mean measurements of the nose in millimeters.

AGE IN YEARS	ANTEROPOSTERIOR LENGTH OF NOSE			NOSE LENGTH (NASION TO TIP OF NOSE)		
	CLEFT PALATE		NORMAL	CLEFT PALATE		NORMAL
	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE
1	16.2	19.5	18.0	33.7	34.1	30.8
3	17.2	21.6	21.4	37.4	38.9	37.0
6	19.8	24.0	23.1	44.5	43.3	41.1

Discussion

The most important finding to emerge from this investigation is that the recent generation of repaired cleft palate patients differs markedly from that of previous years. The conclusions of Graber (4), Buck (2), Snodgrass (8), Krogman (5), and Slaughter and Brodie (7) relative to the deleterious effects of surgery on the growth of the middle face do not apply to the children in our study. The difference in our findings stems from the fact that surgical techniques have changed and the procedures currently in vogue are not inhibitive to the growth process. Since palatal repair for about 50% of our patients was completed before the age of two and one-half years, there is no evidence whatsoever that age at operation, per se, is a critical factor.

The present study further confirms previous reports on mandibular

deficiency in the cleft palate population. Cleft lip and palate emerges all the more as an anomaly complex involving other organs contiguous to the maxilla.

When Graber (4) first reported that the mandible was smaller in the cleft subject, this finding was regarded with some skepticism since he had grouped all clefts into a single population. It was felt that if a number of micrognathic mandibles present in a sample of isolated clefts were included in the population of clefts, the tendency toward mandibular micrognathia would have been exaggerated. Borden (1) analyzed mandibular growth in a population restricted to clefts of the lip and palate and from which all isolated clefts were excluded. Borden's analysis of subjects in our longitu-

TABLE 4. Average soft tissue thickness in millimeters.

AGE IN YEARS	OVERLYING								
	NASION			POINT A			POGONION		
	CLEFT PALATE	NORMAL		CLEFT PALATE	NORMAL		CLEFT PALATE	NORMAL	
	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE	MALE AND FEMALE	MALE	FEMALE
1	11.1	11.2	11.0	7.7	9.8	9.0	10.6	9.0	9.0
3	11.0	10.2	9.7	8.0	12.2	10.7	10.4	10.0	10.1
6	10.5	9.9	9.0	8.6	12.7	11.3	11.5	11.0	10.0

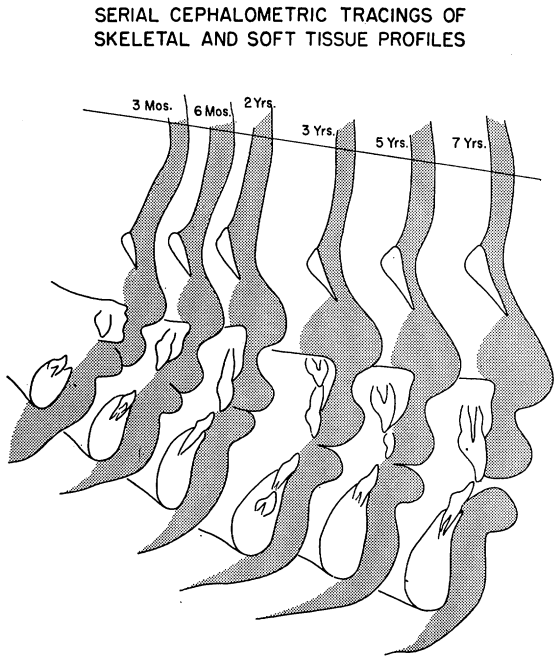


FIGURE 10. Tracings of cephalometric x rays on the same cleft palate individual depicting changes in skeletal and soft tissue profiles over a period of seven years.

dinal study, compiled with our own results, corroborated previous claims that the mandible is indeed smaller in patients with cleft lip and palate.

Although the skeletal profile of the cleft lip and palate population reported herein differs from the normal sample in demonstrating less mandibular prognathism and greater convexity, the difference was not so great as to render the individual strikingly different from what is encountered in the general population of children.

The reduction of the convexity of the soft tissue profile of the cleft population resulted from two factors. If the integument of the nose is considered part of the facial contour, the characteristic flattening of the nasal tip contributes to a reduced facial convexity. In addition, the soft tissue in the subnasale area behaved independently of its skeletal counterpart, point A. The finding that the soft tissue of the upper lip overlying point A is reduced in thickness in the cleft lip sample when compared to normal children of the same age is of special interest (Table 4). This finding may be due to an intrinsic deficiency of soft tissue in this region or as a result of lip surgery, or to a deficiency in the anterior nasal spine. In any event, it became apparent that the soft tissue profile must be appraised not only in terms of its underlying skeletal framework, but also as an independent variable (Figure 10).

Summary

Measurements of the integumental and skeletal profile on 21 subjects with complete unilateral cleft lip and palate were obtained from serial lateral cephalometric x rays extending from three months to seven years of age. The findings were compared with those from a similar study on normal individuals.

The mandible was found to be less prognathic for the cleft sample than for non-clefts. This corroborates previous reports and adds to the concept that cleft lip and palate must be considered as an anomaly complex involving organs contiguous to those cleft.

Convexity of the skeletal profile was greater for the clefts than for the normal population. Despite the fact that at least one half the cases were subjected to palatal repair at two and one-half years of age and less, there was no evidence to indicate that current practice in plastic surgery produced the growth-inhibiting results reported in earlier investigations. The reduction of the convexity of the soft tissue profile of the cleft population resulted from two factors: the characteristic flattening of the tip of the nose, and independent behavior of soft tissue in subnasale area. Another possible consideration is the relative deficiency of the anterior nasal spine and its failure to give skeletal support to the integument in this region.

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References

1. BORDEN, G. H., Mandibular growth in the cleft palate infant. M.S. thesis, Univ. of Illinois, 1953.
2. BUCK, M. W., An x-ray study of cleft palate, oral and pharyngeal structures and their functioning. Ph.D. dissertation, Univ. of Iowa, 1951.
3. DOWNS, W. B., Variations in facial relationships: their significance in treatment and prognosis. *Amer. J. Orthod.*, 34, 812-840, 1948.
4. GRABER, T. M., Craniofacial morphology in cleft palate and cleft lip deformities. *Surg. gyn. Obst.*, 88, 359-369, 1949.
5. KROGMAN, W., The problem of the cleft palate face. *Plastic reconstr. Surg.*, 14, 370-375, 1954.
6. PRUZANSKY, S. and LIS, E., Cephalometric roentgenography of infants—sedation, instrumentation, and research. *Amer. J. Orthod.*, 44, 159-186, 1958.
7. SLAUGHTER, W. B. and BRODIE, A. G., Facial clefts and their surgical management in view of recent research. *Angle Orthod.*, 19, 203-224, 1949.
8. SNODGRASSE, R. M., Heredity and cephalo-facial growth in cleft lip and/or cleft palate patients. *Cleft Palate Bull. Monogr.*, Suppl. 1, 1954.
9. SUBTELNY, J. D., A longitudinal study of soft tissue facial structures and their profile characteristics defined in relation to underlying skeletal structures. *Amer. J. Orthod.*, 45, 481-507, 1959.