# Facial Growth in Children with Isolated Cleft Palate

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Isolated cleft palate differs from cleft lip and associated cleft palate both etiologically and developmentally. There are basic structural differences, especially between complete and degrees of incomplete clefting and these are acted upon by pre- and post-natal environmental influences which are also dissimilar. As a result the morphological differences between cleft types are greater in some respects than between cleft and normal. For this reason a study of craniofacial growth in children with clefts of the lip and palate is only valid when there is separation of individual cleft types and degrees of clefting. Unfortunately much of the information provided in the literature is derived from studies in which several types of cleft were considered together, usually because the samples available were too small to permit subdividing.

Although it has long been observed that cleft palate and subsequent surgical repair result in a smaller maxilla than normal, direct evidence is difficult to obtain.

Coupe and Subtelny (1) found a deficiency of hard palate tissue in unoperated infants with cleft palate. Harvold felt that there was no reduced growth potential associated with cleft palate (2). Mestre and associates (3) and Ortiz-Monasterio (4) found that children with unoperated clefts achieved essentially normal adult faces. Subtelny (5)noted that the maxilla was wider in unoperated infants.

Graber (6), in his study of 175 cleft cases (40 of which were isolated cleft palate), produced measurements from cephalometric radiographs which showed maxillary deficiencies, especially in severe cases where surgery had been performed very early and repeatedly. His sample contained unoperated cases in which growth approximated the normal. Jolleys (7) also noted maxillary underdevelopment in his sample of 94 cases (29 with isolated cleft palate), but it was uncorrelated with the age at which surgery was performed. The extensive operations caused more growth retardation than did the simple operations and Jolleys con-

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cluded that fibrosis of the palate was the major factor. He pointed out that Graber's sample did not represent modern rehabilitative methods. Swanson and associates (8) found that, in their sample of 50 cleft palate cases, the maxilla was retruded (that is, point "A" was more posterior). Ross and Coupe (9), however, concluded from their study of twins that an underdeveloped maxilla in isolated cleft palate was partly a developmental fault.

The mandible in isolated CP has also been the subject of controversy. Many animal studies in which cleft palate is induced experimentally have noted a short mandible as one frequently associated anomaly (10, 11). Borden (12) noted that, in infants with cleft palate, mandibular growth followed the normal pattern but the mandibles were smaller or had a retarded growth expression.

Pruzansky and Richmond (13) found that even in Pierre-Robin syndrome the micrognathic mandible is capable of proportionately adequate growth which eventually reduces the retrognathic profile. Graber (12)noted that the mandible was retrusive in cleft palate and suggested mandibular underdevelopment. Swanson and associates (8) found that the chin was retrusive compared to Down's "ideal" normals, but not to their own randomly chosen sample of nonclefts. They pointed out that retrusion may be the result of altered position rather than underdevelopment. In unoperated cases, they found that the mandibles were similar to controls.

Ross and Coupe (9) found the mandible to be smaller in isolated cleft palate and the gonial area so altered that there is a steep mandibular plane, obtuse gonial angle and decreased posterior facial height. They concluded that the mandible was developmentally smaller, but the altered position and therefore the angular measurements could be the result of secondary environmental influences including surgery. An increase in mandibular width was also noted. A review of many of the relevant studies has recently been published (14).

It has generally been noted that jaw relations have been reasonably good in isolated cleft palate. Levin (15) found in a survey of 847 subjects with all types of clefts that 11.7% of the isolated soft palate clefts had a positive ANB angle (that is, a concave type of face) while 19.7% of the hard and soft palate clefts showed this characteristic. Johnston (16) felt that growth was not markedly affected and the deformity was neither great nor difficult to treat orthodontically. Ross and Coupe (9) found that the overall facial rotation and retrusion resulted in acceptable profiles and skeletal relations. Swanson and associates (8) found that the skeletal profile very closely approximated the controls. They noted that 80% of the isolated cleft palate cases were in neutrocclusion. An excess freeway has been found in many cases, which Graber felt was due to maxillary deficiency related to palate surgery (2). However, Ross and Coupe (9) found that in many cases the mandibular arch is as likely to be responsible for a lack of adequate vertical eruption of the teeth.

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Altered jaw growth and development have been of primary concern to the orthodontist, but other differences have been reported. These children are smaller (9, 17), probably have a delayed maturation (18), and have a higher incidence of other congenital anomalies (19). The morphological aberrations in the head region seem to be confined to the jaws and nasal septum; the lateral facial bones (4) and cranial base (9, 17) are normal and unaffected by the presence of a cleft. Subtelny (20) has published a review of these and other growth studies.

The present study was designed to accomplish two aims: 1) to determine the difference in craniofacial morphology between normal children and children with cleft palate at six years of age, and 2) to discover whether the observed differences remained constant, became more pronounced, or were lessened through compensatory growth.

## Sample

Children with severe clefting of both the hard and soft palates were chosen for this study. (In our clinic, "severe" indicates that the cleft extends close to the incisive foramen.) Additional criteria were: all were Caucasians, with no known additional congenital anomalies, with reasonable dentitions (that is, not multilated by extractions), and had a successful palatoplasty (only four cases had more than one operation). The mean age of palatal surgery for the group was 26 months. Four of these children had been diagnosed as having Pierre-Robin syndrome at birth. None were undergoing orthodontic therapy during the period of observation.

Part I of the study used 30 children (15 males and 15 females) with cleft palate (as defined above). The mean age of this group was 6.0 years with a range of 5.4 to 7.0 years. A control group of 30 six-year-old noncleft children (15 males and 15 females) was randomly selected from the Burlington Orthodontic Research Center (Table 1).

	cleft palate					control			
	mean age and range	male	female	total	male	female	total		
Part I	6.0(5.4-7.0)	15	15	30	15	15	30		
Part II									
Group A	6.3 (5.4-6.8)	9	7	16					
(6-9 years)	8.8 (8.0-9.7)								
Group B	9.3 (8.2 - 9.8)	11	8	19					
(9-12 Years)	11.8 (10.8–13.2)				15	15	30		
Group C	11.9 (11.0-13.2)	6	8	14					
(12–15 Years)	15.0 (14.2-15.8)								

TABLE 1. Age and sex distribution of cleft palate sample and controls.

The sample for part II of the study (Table I) consisted of three groups of cleft palate children with serial records from age 6 to 9 years (Group A), age 9 to 12 years (Group B), and age 12 to 15 years (Group C). In this way three consecutive serial studies provided a complete range of growth from 6 years to 15 years. Records of 36 individuals were reviewed for the study. A control group of 30 children with serial records at age 6, 9, 12, and 15 years was randomly obtained from the Burlington Orthodontic Study. Since the Burlington records were taken on or about the child's birthday, the range is negligible at each age.

# Method

Cephalometric radiographs in the lateral view were used to locate the landmarks shown in Figure 1. Individual tracings were made and used for direct measurement and for the construction of average facial diagrams.

The following measurements were recorded for all subjects: cranial base length, nasion to basion (N-Ba); maxillary length, anterior nasal spine to posterior maxilla (ANS-PM) (PM is constructed as the intersect of palatal plane and a perpendicular from the palatal plane to the pterygo maxillary fissure); mandibular length, greatest length of mandible between two points approximating gnathion and condylion (Gn-Con); anterior facial height, nasion to menton (N-Men) projected on the facial plane (N-Pog); nasal height, nasion to anterior nasal spine (N-ANS) projected on the facial plane (N-Pog); oral height, anterior nasal spine to menton (ANS-Men) projected on the facial plane (N-Pog); posterior facial height, sella to gonion (S-Gon) projected on the facial plane (N-Pog); craniofacial angle, relation of facial profile to cranial base (N Ba-N Pog); maxillary inclination, angle of maxillary plane to cranial base (N Ba to ANS-PM); mandibular inclination, angle of mandibular plane to cranial base (N Ba-Md pl); profile jaw-relations, represented by the prognathism of point A (on the maxilla) and point B (on the mandible) relative to nasion, using the A-N-B angle; gonial angle; and interincisal angle.

It was known that children with cleft palate would probably be smaller children on the average (9, 17) and that cranial base length accurately reflects general body size (17). The diagrams were therefore enlarged to a fixed cranial base length, eliminating this variable which could be misleading in interpreting the diagrams. The linear measurements were also size-adjusted (Table 2).

When analyzing the growth trends, it was desirable to avoid direct comparison of linear or angular measurements, since the smaller samples and size differences might permit misleading conclusions. Instead, the growth changes were noted and expressed as either angular changes (Table 3) or as percentage increments (Table 4).



FIGURE 1. Tracing of a lateral cephalometric radiograph indicating the landmarks used in the study: N, nasion; ANS, anterior nasal spine; A, point "A" on maxilla; B, point "B" on mandible; 1, maxillary and mandibular central incisors; Pog, pogonion, Gn, gnathion; Men, menton; Gon, gonion; Con, condylion; S, sella; Ba, basion; Ptm, pterygo-maxillary fissure; KR, key ridge of zygoma; PM, posterior of maxilla. The heavy lines indicate the manner in which the average facial diagram related to the tracing.

## Findings

Part I of the study was an assessment of craniofacial morphology at age six years. The major findings are presented in Table 2. Figure 2 shows the average facial diagrams of the cleft palate and control groups size-adjusted and superimposed on the cranial base (2A) and on the facial profile (2B).

Part II of the study was an evaluation of the direction and extent of facial growth from age 6 years to 15 years. The major findings are presented in Tables 3 and 4. In Table 3, the absolute increments of growth are expressed as a percentage of the measurement at the beginning of TABLE 2. Measurements on 30 six-year-old cleft palate children compared with 30 six-year-old controls. The mean linear values for the cleft palate group were adjusted on the basis of the cranial base length. This compensates for the overall body size differences between the groups. The standard deviation and t value are given. t values with one asterisk are significant at the .01% level, those with two asterisks are significant at the .05% level.

	c	left palat	e	contro		
measurement	actual mean	SD	adjusted mean	mean	SD	t
linear						
cranial base length	$94.7 \mathrm{mm}$	4.35	$95.5 \mathrm{mm}$	$95.5 \mathrm{mm}$	3.49	0.797
maxillary length	$47.3 \mathrm{mm}$	2.86	$47.9\mathrm{mm}$	$51.0\mathrm{mm}$	2.26	$4.919^{**}$
mandibular length	$94.8 \mathrm{mm}$	5.30	$95.6 \mathrm{mm}$	$96.5 \mathrm{mm}$	3.98	0.727
anterior facial ht	$104.1 \mathrm{mm}$	5.50	$105.0 \mathrm{mm}$	$101.5 \mathrm{mm}$	5.47	2.461*
nasal ht	$43.5 \mathrm{mm}$	2.36	$43.9\mathrm{mm}$	$43.0 \mathrm{mm}$	2.13	1.540
oral ht	60.6mm	4.30	$61.1 \mathrm{mm}$	$58.5\mathrm{mm}$	4.54	$2.295^{*}$
posterior facial ht	61.4mm	2.94	61.9mm	$63.5 \mathrm{mm}$	4.17	1.711
angular						
craniofacial angle	55.3°	5.65		58.2°	2.62	2.542*
maxillary inclina-			-			
tion	29.8°	3.41		24.4°	2.37	7.101**
mandibular inclina-						
$ ext{tion} \dots \dots \dots \dots$	57.3°	4.53		$51.5^{\circ}$	4.36	5.038**
gonial angle	133.7°	5.48		130.2°	4.21	2.767**
profile jaw-rela-						
tions	3.7°	2.51		4.6°	2.10	1.500
interincisal angle	154.3°	11.92		144.5°	11.64	$3.215^{**}$

\* .01 < P < .05

\*\* P < .01

each age span. It should be noted, therefore, that one millimeter of growth at age 6 will produce a higher percentage increment than would a millimeter at age 12. In Figure 3, both the direction and extent of movement of the major facial landmarks relative to the cranial base are illustrated by arrows.

#### Discussion

PART I: ANALYSIS AT 6 YEARS. There are major differences in craniofacial morphology between a child with a cleft palate and a normal child, but at present it is difficult to attribute these differences to either congenital or postnatal influences. Certainly there are intrinsic factors related to genotype or embryonic development, as well as extrinsic factors related to intrauterine environment, postnatal environment, function, and surgical treatment. When a group of six-year-old children is investigated, all of these factors have been influential during a period of

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		cleft palate		control		means			
measuremeni	age	male	female	male	e female	cleft		control	
cranial base length	6–9	6.9	4.7	4.7	4.9	5.8		4.8	
	9-12	4.6	4.0	4.2	4.6	4.3	14.7	4.4	12.3
	12 - 15	5.8	3.4	4.3	1.9	4.6		3.1	
maxillary length	6–9	3.4	5.0	6.7	5.3	4.2		6.0	
	9-12	3.9	6.5	4.4	6.4	5.2	13.6	5.4	16.3
	12 - 15	5.2	3.2	6.1	3.7	4.2		4.9	
mandibular length	6-9	6.9	6.4	8.6	7.6	6.7		8.1	
	9-12	6.5	6.9	6.6	9.0	6.7	20.0	7.8	22.5
	12 - 15	6.9	6.3	8.7	4.5	6.6		6.6	
anterior facial	6-9	4.3	5.3	7.2	6.6	4.8		6.9	
height	9 - 12	6.1	6.2	6.9	6.9	6.2	17.5	6.5	18.5
	12 - 15	8.0	5.0	6.6	3.5	6.5		5.1	
oral height	6–9	4.3	4.7	6.3	5.5	4.5		5.9	
	9 - 12	6.8	9.5	5.4	7.6	8.2	20.9	6.5	17.3
	12 - 15	9.3	7.1	6.8	2.9	8.2		4.9	
posterior facial	6–9	6.9	6.5	7.8	8.4	6.7		8.1	
height	9 - 12	7.5	6.1	6.9	10.0	6.8	21.8	8.5	23.4
-	12 - 15	9.8	6.8	9.0	4.6	8.3		6.8	

TABLE 3. Growth increments at each age period expressed as a percentage of the initial measurement for that period. For example, the cranial base length increased in the male cleft group from age 9 to 12 by 4.6% of its length at 9 years.

extremely rapid growth. Therefore no conclusions can be drawn from this study as to the causes of the peculiar morphological features identified.

At age 6 years, the entire face of the child with a cleft palate appears to be rotated posteriorly relative to the midline cranial base (Figure 2A). Since the cranial base is normal in this condition (9, 17), the conclusion is inescapable that the forward growth of the face was deficient. This is understandable when considering the maxillary area, since all the inhibiting factors act on this complex of bones. The maxillary length measurements were significantly less (Table 2), although the posterior limit of the maxilla was not altered (Figure 2A), which indicated that pharyngeal patency was maintained.

It is less obvious why the mandible should be deficient in anterior growth. The data reveal that the mandible was of normal length (Table 2) but the chin was posteriorly displaced (Figure 2A). This retropositioning of the chin was essentially the result of mandibular rotation with subsequent remodeling of the muscle attachments in the gonial area (indicated by the increased gonial angle and mandibular inclination, Table 2) and may have been a functional response to the altered maxillary complex. The normal mandible and tongue established a satisfactory relation with a small, shallow-vaulted, maxilla. The changes may also have been induced by mouth breathing, a common finding.

		by p	eriod	net change		
measurement	age	cleft	control	cleft	control	
craniofacial angle	6–9	-0.8	0.7			
	9 - 12	0.4	1.2	-0.3	3.1	
	12 - 15	0.1	1.2			
maxillary inclination	6-9	0.4	0.6			
5	9 - 12	-0.1	0.6	1.0	1.2	
	12 - 15	0.7	0.0			
mandibular inclination	6-9	-0.4	-0.3			
	9 - 12	0.2	-0.8	-0.1	-2.3	
	12 - 15	0.1	-1.2			
gonial angle	6-9	-2.0	-1.8			
gomm ungeo	9 - 12	-0.6	-1.4	-3.9	-4.7	
	12 - 15	-1.3	-1.5			
profile jaw-relations	6-9	-1.1	-0.5			
promo Jan record	9-12	-1.0	-0.7	-2.2	-2.0	
	12 - 15	-0.1	-0.8			
interincisal angle	6-9	-9.9	-14.3			
mitor monotal tanglo	9-12	-3.3	-0.5	-11.8	-14.5	
	12 - 15	1.4	0.3			
			1		[	

TABLE 4. The change, in degrees, that occurred in each angle during the age span indicated is given. The net change is a total of the changes from 6 years to 15 years.



FIGURE 2. Mean facial diagrams for the six-year-old cleft palate group (interrupted line) and control group (solid line) superimposed on the cranial base, A, and on the facial plane, B.

Thus despite the greater anterior-posterior deficiency of the maxilla as compared to the mandible, the profile jaw-relations were not significantly altered (Table 2 and Figure 2B). This is important for two reasons; first, from outward appearances the facial profiles were normal;



FIGURE 3. The direction and extent of growth from age 6 years to 15 years of some of the facial landmarks are indicated. Growth increments were plotted from the base line six-year-old groups and represented by arrows.

and second, the basal bones of the two jaws were close to a normal anteroposterior relationship, so that in this respect the teeth could occlude normally.

The maxillary incisors were positioned several millimeters more posteriorly than in the normal child (Figure 2B). This was mainly due to the small maxilla, but in addition the incisors were slightly retruded relative to the basal bone. This may be an inhibition of normal downward and forward dental eruption because of scar tissue in the palate into which peridontal fibers from the teeth are inserted. Another possibility is the presence of hypertonic lip and cheek musculature, a common clinical finding in many cleft palate children. Whatever the cause, a reasonable incisal relationship, but with greater-than-normal interincisal angle, was found.

As a result of the change in mandibular position, the anterior vertical height of the face was increased in cleft palate, with excessive oral height (Table 2 and Figure 2B). Interpretation of the posterior vertical dimensions depends on whether one considers relations to the cranial base (in which case the maxilla is markedly deficient and the mandible only slightly deficient, Figure 2A) or relations to the face ( in which case the maxilla is only slightly deficient, the mandible slightly excessive, Figure 2B).

Since the oral vertical height in the dental area was excessive, and since it would be expected that eruption of the maxillary buccal teeth would be inhibited, it would thus be expected that the occlusal plane would be located superiorly in the cleft group, with overeruption of the mandibular buccal teeth to compensate. Inexplicably, the position of the occlusal plane indicated a normal mandibular eruption but an excessive maxillary eruption (Figures 2A and 2B). This confirms the findings of Ross and Coupe in monozygotic twins (9). The clinical implication of this finding is that, when there is an excess freeway space due to lack of dentoalveolar vertical development, it should not be assumed that the deficiency is in the maxillary arch but is quite likely to be in the mandibular arch.

PART II: GROWTH TRENDS FROM 6-15 YEARS. The absolute difference in cranial base length (which is related to the size of the child) at six years was maintained, because of a slightly larger percentage growth increment in the children with cleft palate (Table 3).

The tendency in a normal child is for the face to emerge from under the cranial base slightly (that is, the craniofacial angle increases, Table 4). This tendency did not occur in the cleft palate group. The retruded face in cleft palate became relatively more retruded.

Before discussing the growth of the jaws in detail, two things should be noted. First, the child with a cleft palate seems to mature later than his noncleft contemporaries. This has been the subject of a preliminary investigation by Menius and associates (18) and is the clinical impression of many who deal with these children. Second, male and female growth patterns follow different schedules. Normally, the prepubertal period of accelerated growth, often termed the "growth spurt", occurs later in the male, and continues longer, especially for the mandible. These two factors must be taken into account in the evaluation of the results in Table 3.

The maxillae of the children with cleft palate grew proportionately slightly less in length than the controls, indicating an increasing retardation in absolute length (Table 3). Maxillary inclination remained constant in both groups (Table 4).

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Mandibular growth analysis requires more interpretation (Table 3). The female control group had a large growth increment from 9-12 years, but subsequently the increment was very small, indicating that most of the girls had had their pubertal spurt prior to age 12, with growth rapidly subsiding after this. The cleft palate females had a less dramatic increase from 9-12 years, but maintained this level in the years from 12-15, indicating that a large number of these girls had their growth spurt after the age of 12. Predictably, the males in the control group had the greatest increment in the period from 12-15 years. The cleft palate males, however, showed only a slight increase in the 12-15 year increment, indicating either that they do not have a growth spurt, or that many of these boys have their greatest mandibular growth after the age of 15 years. We believe the latter to be the case. If the study could have been continued a few additional years, the results might have shown that the cleft palate groups continued their incremental growth and that the end result would be mandibles of a similar size in both groups. In the present findings, however, the mandible in children with cleft palate was found to grow less from 6–15 years.

The inclination of the mandible remained essentially unchanged in the cleft palate groups, although the gonial angle became more acute (Table 4). In the control group there was a tendency towards a reduction in angulation, and a corresponding reduction in the gonial angle.

Profile jaw-relations remained consistent in both groups, with a mild trend towards a less convex face with age.

Anterior facial height, which was greatest at 6 years in the cleft group, showed smaller increments from 6-9 years. The earlier growth spurt of the female controls increased their facial height more from 9-12 years, but growth decreased markedly after 12 years. Oral height showed a similar pattern, with large growth increments in the cleft groups. This difference in oral height was the greatest difference in any measurement between the cleft and the control groups.

Posterior facial height showed less growth in the cleft group from 6–9 years (Table 3), with approximately equal growth thereafter, the observed mean differences being related to the age at which accelerated growth occurred.

The height differences might explain the high frequency of anterior open bites appearing in the permanent dentition of children with cleft palate.

The interincisal angle indicates that most of the uprighting of the incisors occurred between the ages of 6 and 9, with some further uprighting in the cleft palate group from 9-12 years.

Figure 3 illustrates both the direction and extent of movement of the major facial landmarks relative to the cranial base. The overall impression is that the cleft group grew less and in a more vertical direction, especially the maxilla and its directly associated structures. Of particular

interest to orthodontists are the movements of points "A" and "B", clinically the most significant landmarks. While the normal points "A" and "B" followed the same pattern as the basal maxilla (ANS) and mandible (Pogonion), in the cleft palate group they did not grow forward with the basal bone to the same extent. This may be the restricting effect of the palatal scar tissue on the maxillary dentoalveolar structures, or tight musculature, together with the secondary retrusion of the mandibular incisors.

## Summary

Cephalometric radiographs were used to determine the facial characteristics of 30 children with isolated cleft palate at age 6 years as compared with 30 noncleft children. Serial radiographs were used to establish subsequent growth trends in both groups to age 15 years. There was evidence of progressive maxillary underdevelopment, but with acceptable facial balance due to positional changes of the mandible.

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