Midfacial Morphology in Adult Unoperated Complete Unilateral Cleft Lip and Palate Patients

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ABSTRACT

Objective: To examine lateral cephalometric radiographs of adult unoperated cleft lip and palate patients for the purpose of clarifying whether maxillary deficiencies observed in treated cleft patients result from intrinsic defects or surgical intervention early in life.

Materials and Methods: This retrospective study examined lateral cephalograms of 30 adult patients with nonsyndromic complete unilateral cleft lip and palate (CUCLP). The lateral cephalograms were traced and evaluated for size and position of the cranial base, maxilla, maxillary dentition, mandible, and mandibular dentition as well as for vertical relationships. Comparisons with 30 adult noncleft individuals were made.

Results: In unoperated adult cleft lip and palate patients, the cranial base angle was increased with the anterior cranial base reduced in length. The maxilla was found to be normal in size and somewhat prognathic in position. Both the maxillary and mandibular incisors were relatively upright. The mandible was smaller in size and posteriorly positioned.

Conclusions: The potential for normal growth of the maxilla exists in patients with CUCLP. It is likely that disturbances of maxillary growth in surgically operated cleft patients are related primarily to the surgical intervention.

KEY WORDS: Cleft lip and palate; Cephalometrics; Unoperated; Adults

INTRODUCTION

Inhibition of the growth and development of the nasomaxillary complex in treated cleft lip and palate patients is an extensively debated topic. Graber[1] in his landmark study documented severe three-dimensional (3-D) maxillary collapse in patients with complete cleft lip and palate after surgery.

These patients often present with anterior and posterior crossbites and midface deficiency with a tendency toward Class III malocclusion. Williams et al[2] examined repaired cleft patients at 5 and 12 years of age and observed that 40% of the patients at 5 years of age had poor dental arch form and 70% at 12 years of age had midfacial retrusion. The abnormal facial morphology in treated cleft patients has been attributed to two factors—intrinsic developmental deficiency or iatrogenic factors introduced by treatment.

Bishara,[3] Isiekwe and Sowemimo,[4] and Yoshida and Nakamura[5] claim that maxillary deficiency in cleft individuals is an intrinsic primary defect. However, Ortiz-Monasterio et al,[6] Bishara et al,[7] Mars and Houston,[8] and Capelozza et al[9] have written that maxillary deficiency is secondary to surgical repair. If maxillary deficiency is a complication of the surgical intervention, it is important to determine optimal conditions for palatal and lip closure in terms of timing and the choice of procedure. Examining unoperated adult patients with cleft lip and palate will help elucidate the effects of surgery on the nasomaxillary complex.

In developed countries, most patients with cleft lip and palate undergo surgery early in life, thereby eliminating the opportunity to observe and study large numbers of patients with unrepaired clefts at the later stages of development. To obtain information regarding significant populations of adults with unoperated cleft lip and palate, data can be collected from regions where early surgery is not readily available, such as remote areas of India (Figures 1 through 3).

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FIGURE 1. Extraoral photographs of a 26-year-old, male, adult unoperated complete unilateral cleft lip and palate patient. (A) Right Profile. (B) Frontal. (C) Left Profile.

The craniofacial growth in cleft lip and palate is influenced by the inherent genetic growth potential, the functional abnormalities due to cleft, and the surgical intervention performed to repair the cleft. The genetically determined components of craniofacial growth in cleft patients can be separated from surgical effects by investigating an unoperated sample. Hence, it was decided to conduct a study using lateral cephalometric radiographs to evaluate maxillary and mandibular morphology and to clarify whether the maxillary deficiency seen in operated cleft palate patients is a product of intrinsic defects or iatrogenic interferences.

The objective of this study is to examine lateral cephalometric radiographs of adult unoperated cleft lip and palate patients and to determine whether maxillary deficiencies observed in the cleft patients result from intrinsic defects or surgical intervention early in life.

MATERIALS AND METHODS

This retrospective study evaluated lateral cephalograms of 30 adult patients of Indian origin from Karnataka state, who had an untreated complete unilateral cleft lip and palate (CUCLP). Only nonsyndromic clefts were included in this study. All the patients were over 18 years of age at the time of records. The sample consisted of 22 male and eight female patients. The control group consists of 30 lateral cephalograms of normal geographically similar Indian individuals, 15 males and females with Class I molar relationship, normal overjet, and overbite with full complement of teeth present and ranging in age between 18 and 25 years. These data were previously collected to establish cephalometric norms for the Indian population.

The lateral cephalograms were obtained for the cleft patients and the normal controls using a fixed subject to x-ray distance of 5 feet. The head was oriented with the FH plane parallel to the floor, with teeth in centric occlusion and lips lightly in contact. The machine was calibrated for 8% magnification, and this magnification was not corrected in our measurements.

Soft tissue and bony structures were traced on polyester film, and the mean shadow of bilateral structures was traced to minimize slight errors in positioning or skeletal or dental asymmetries. The following landmarks were identified on each cephalogram (Figure 4):

- A point (A), B point (B), Anterior nasal spine (ANS), Basion (Ba), Condylion (Co), Gnathion (Gn), Gonion (Go), Menton (Me), Nasion (Na), Orbitale (Or), Pogonion (Pog), Porion (Por), and Sella (S).

The following linear and angular measurements were used in this study:

- Cranial base: N-S-Ba (°), N-S (mm), N-Ba (mm), and S-Ba (mm).
- Maxillary skeletal: S-N-A (°), Point A to Na perpendicular (mm) (the distance from Point A to a line through nasion that is perpendicular to the Frankfort horizontal), Co-A (mm), and NPog-A (mm).
- Maxillary dental: upper incisor (U1) to SN (°), U1 to NA (mm), U1 to NA (°), and U1 to A vertical (mm).
- Mandibular dental: lower incisor (L1) to mandibular plane (°), L1 to NB (°), L1 to NB (mm), and L1 to APog (mm).

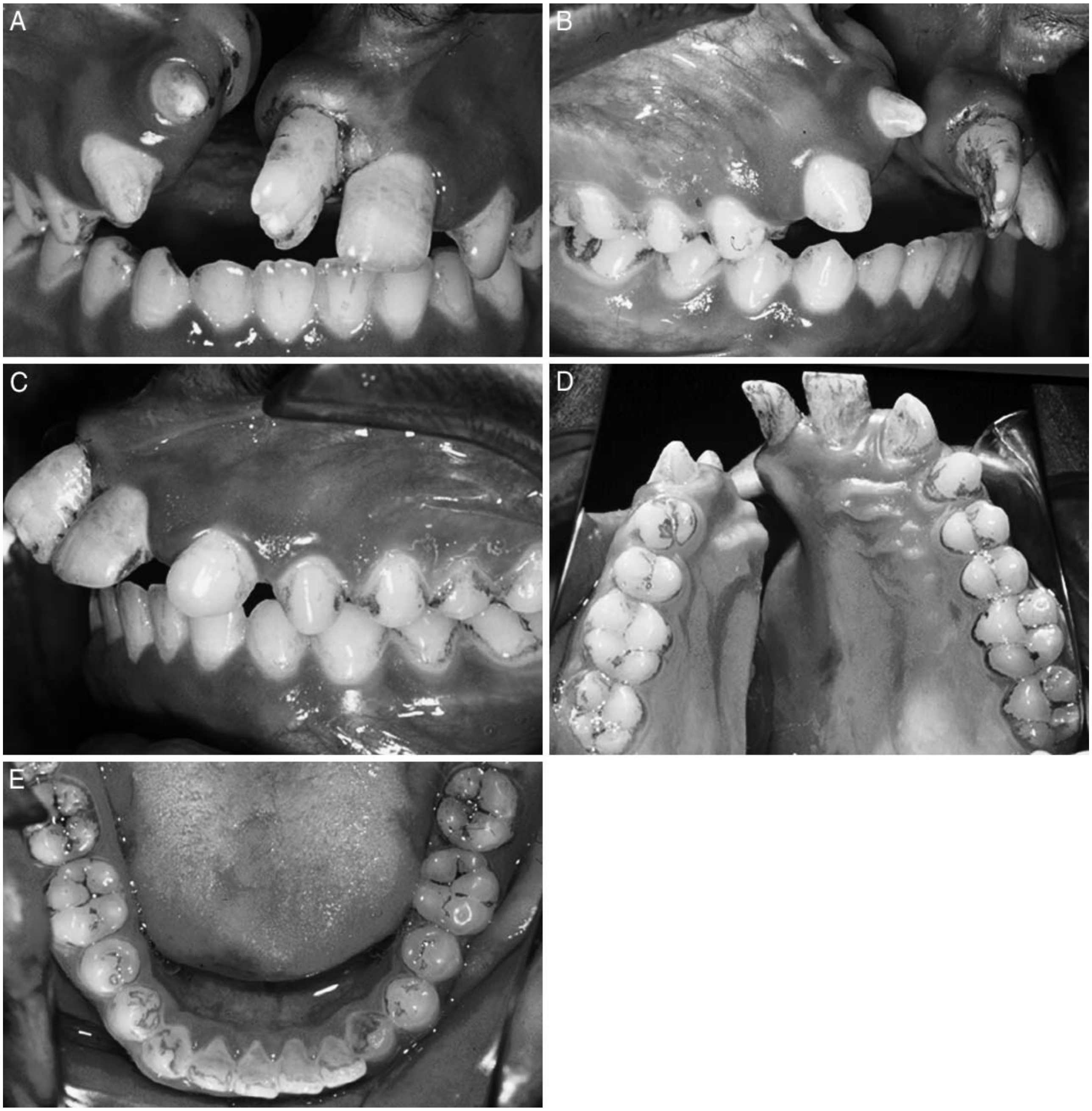

FIGURE 2. Intraoral photographs of the same 26-year-old, male, adult unoperated complete unilateral cleft lip and palate patient. (A) Frontal. (B) Right Buccal. (C) Left Buccal. (D) Maxillary Occlusal. (E) Mandibular Occlusal.

- Mandibular skeletal: S-N-B (°), facial angle N-A-Pog (°), Pog to Na perpendicular (mm) (the distance from pogonion to a line through nasion that is perpendicular to Frankfort horizontal), and Co-Gn (mm).
- Vertical: Ba-Na-Go (°), Frankfort mandibular plane angle FMA (°), lower anterior facial height LAFH (mm), upper to lower anterior facial height ratio and posterior facial height to anterior facial height Jaraback ratio (%).

All the data were analyzed statistically for means and standard deviations. The mean values for each parameter in both the groups were then compared using the student's *t*-test.

RESULTS

The mean values, standard deviations, and student's *t*-tests of all the variables used in this study are shown in Table 1. The level of significance adopted for statistical test was $P < .05$.

FIGURE 3. Lateral Cephalogram of the same 26-year-old, male, adult unoperated complete unilateral cleft lip and palate patient.

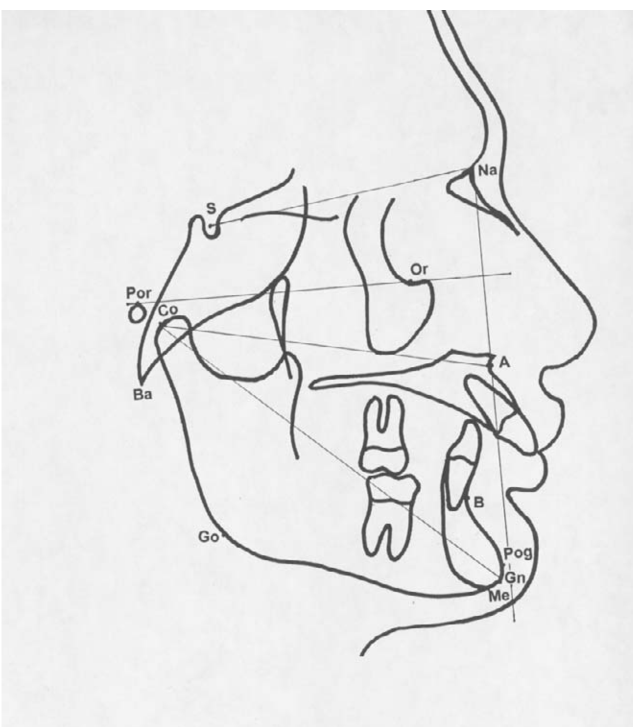

FIGURE 4. Landmarks and lines used in the cephalometric analysis.

Cranial Base

The cranial base angle (N-S-Ba) in unoperated unilateral complete cleft lip and palate CUCLP patients was significantly larger by 5.6° when compared with the normal control. Anterior cranial base length (N-S) was significantly smaller by 2.96 mm and S-Ba as well as N-Ba were similar to the noncleft control values.

Maxillary Skeletal

The SNA angle of 84.5° was larger by 1.85° than the normal control. Point A was significantly forward by 2.48 mm when measured from nasion perpendicular and 3.16 mm when measured from the nasion-pogonion line. The effective length of the maxilla measured from condylion to point A was similar to the control.

Maxillary and Mandibular Dental

All the variables evaluated for the maxillary and mandibular dentition showed that the incisors were more retroclined as well as retrusive when compared with normal control values.

Mandibular Skeletal

The mandible in adult unoperated CUCLP was smaller in dimension and positioned more posteriorly than in control subjects. This was indicated by a reduced SNB angle by 2.9° and Co-Gn length reduced by 4.45 mm.

Vertical Relationship

All patients have some vertical growth history. This sample shows excessive growth when compared with the noncleft sample. The FMA was increased by 5.6° and LAFH by 6.65 mm when compared with normal controls.

The maxillomandibular skeletal relationship suggested a strong Class II skeletal pattern, indicated by an ANB of 7.5°, a 4.75° increase when compared with the control value. This finding was related both to prognathism of the maxilla and retrognathism of the mandible.

DISCUSSION

This study attempts to identify causes of the abnormal craniofacial growth in general and the maxilla in particular in surgically treated cleft lip and palate patients. An examination of unoperated adult complete cleft lip and palate patients eliminates surgery as a cause of any abnormal growth observed and helps determine whether the abnormal growth pattern observed in treated cleft patients is an intrinsic defect or a side effect of treatment.

Table 1. Linear and Angular Measurements of Control and Unoperated Cleft Subjects

Variables	Normal Control (n = 30)		Unoperated Cleft (n = 30)		Diff	*t*-Test
	Mean	SD	Mean	SD		
Cranial Base						
N-S-Ba (°)	127.93	5.77	133.53	5.46	5.6	0.0032*
N-S (mm)	73.76	3.29	70.8	3.88	−2.96	0.0025*
S-Ba (mm)	46.1	5.13	46.51	3.35	0.41	0.7183
N-Ba (mm)	109.28	6.13	111.2	5.98	1.91	0.2486
Maxillary Skeletal						
S-N-A (°)	82.6	3.71	84.5	5.64	1.85	0.1504
Na p A (mm)	−0.56	3.32	1.91	3.37	2.48	0.0050*
Co-A (mm)	95.31	5.65	93.8	5.52	−1.51	0.2665
Npog-A (mm)	1.75	2.36	4.91	5.26	3.16	0.0039*
Maxillary Dental						
U1-SN (°)	112.61	6.71	111.3	8.75	1.31	0.4939
U1-NA (°)	29.85	7.05	26	6.86	−3.85	0.033*
U1-NA (mm)	5.88	2.93	5.53	3.21	−0.35	0.6550
U1-A (mm)	6.83	1.92	5.95	2.82	−0.88	0.1073
Mandibular Dental						
IMPA (°)	104.18	7.25	96.7	9.5	−7.48	0.0058*
L1-NB (°)	29.9	6.47	23.91	8.52	−5.58	0.0065*
L1 NB (mm)	6.25	2.04	5.96	3.42	−0.28	0.7069
L1-Apog (mm)	4.03	2.63	0.65	2.9	−3.38	4.33E-05*
Mandibular Skeletal						
SNB (°)	79.85	3.15	76.95	3.98	−2.9	0.0035*
Facial (°)	87.18	2.76	84.95	4.41	−2.23	0.0341*
NapPog (mm)	−4.4	4.54	−6.08	8.76	−1.68	0.4011
Co-Gn (mm)	120.15	7.35	115.7	7.04	−4.45	0.0165*
Vertical Relationship						
Ba-NA-Go	91.21	3.57	88.96	4.71	−2.25	0.0594
FMA (°)	19.48	4.88	25.08	6.99	5.6	0.0013*
LAFH (mm)	63.96	5.52	70.76	6.65	6.8	0.0002*
UFH/LFH	0.83	0.08	0.69	0.06	−0.14	5.52E-08*
Jaraback %	69.49	6.19	64.95	5.63	4.53	0.0024*
Max Man						
ANB (°)	2.75	2.44	7.5	4.59	4.752	8.23E-06*

* Statistically significant at $P < .05$

Attempts have been made in the past to study the growth pattern in unoperated cleft patients, and there have been conflicting opinions. Some studies combined different cleft types,[4] included patients with lip repair,[6,10] compared cleft patients with normal patients from different racial groups,[11] or used a sample with an extended age group.[8] The study reported in this article is unusual because both the untreated cleft and normal individuals were from the same geographic region and very similar backgrounds; the high suitability of the controls is an important advantage in a study of oral clefts because variability is more likely to be related to the issues being considered.

The results of our study show differences in the measurements of the craniofacial structure between unoperated adults with CUCLP and normal controls. The morphology of craniofacial structures in unoperated cleft patients is more favorable than that observed in surgically treated cleft patients, indicating that surgical intervention interferes with growth processes in cleft patients.

In our study of unoperated CUCLP subjects, the cranial base angle N-S-Ba was larger by 5.6° when compared with the normal control. Contrary to our findings, Mars and Houston[8] and Capelozza et al[9] noted no difference in the cranial base angle, whereas Bishara et al[12] observed reduced cranial base angle in unoperated cleft patients. But, in a surgically repaired cleft lip and palate sample, Ross[13] and Dahl[10] observed a larger cranial base angle, whereas Harris[14] observed reduced cranial base angle. The effect of cranial base angle on the pharyngeal space and the position of the mandible needs further investigation because this may directly affect the breathing pattern. The anterior cranial base length N-S was found to be smaller by 2.96 mm in the cleft group than in the controls, but the S-

Ba and N-Ba lengths were comparable with the controls.

It is of interest to consider whether the shortness of the anterior cranial base is an intrinsic genetic condition or whether the shortness is due to altered growth related to changes in function due to the cleft. Cranial base cartilage was thought to be regulated by the genes, but Wang and Mao[15] recently demonstrated that dynamic forces delivered to the premaxillae of growing rabbits for only 20 minutes per day for 12 days resulted in chondrocyte proliferation in the sphenooccipital synchondrosis. The difference in cranial base length observed in this study could theoretically be related to the altered forces of mastication or swallowing.

The increased SNA angle in unoperated CUCLP subjects can be explained geometrically by protrusiveness of A point and the surrounding dentoalveolar process or possibly by retrusiveness of N because of the short anterior cranial base length in comparison with normal noncleft controls (Figure 3). We are interpreting the finding as maxillary protrusion because usually the position of the maxilla is compared with the anterior cranial base. It should also be noted that the A point is located on the noncleft side. The anterior position of the maxilla on the cleft side is difficult to assess on lateral cephalogram and would need 3-D CT study.

Similar findings of maxillary protrusion were noted by Mars and Houston[8] and Capelozza et al,[9] but these differed from the findings of Isiekwe and Sowemimo[4] and Yoshida and Nakamura.[5] Lambrecht et al[16] suggested that the more protrusive position of the maxilla may be caused by anterior tongue thrusts when the patient is trying to close the cleft while eating or speaking. It can be further concluded that the midfacial hypoplasia observed in repaired CUCLP patients may be the outcome of surgical intervention. In surgically treated patients, inhibition of the normal development of the maxilla has been attributed to scar tissue.[17] There may be no damage to the bone itself because of surgery, but the fibrous scar tissue formed near the bony growth sites may prevent normal maxillary remodeling and development in a downward and forward direction. The extent of the interference will be directly related to the severity of the cleft because more extensive procedures have to be performed to mobilize tissue to close a large defect.

In the past some clinicians have tried to delay the closure of the palate to have a less deleterious effect on the growth.[18] However, the proponents of early closure say that delaying closure of the palate will definitely affect the development of speech and it is not a wise decision to trade-off speech development to improve the growth of the maxilla. Caution should be exercised in interpreting the lateral cephalometric data for the maxillary position because very little information is available on the left- and right-side parasagittaly maxillary deformity.[19]

Anterior crossbite is a common finding in patients with repaired clefts. In our unoperated cleft lip and palate sample, the maxillary incisor position was found to be very similar to that of the control. A tight lip repair may retrocline maxillary incisors but does not have any significant effect on the growth of the midface, as evaluated by Mars and Houston.[8,20] The mandibular dentition was more upright when compared with the normal control possibly because of lower lip pressure exerted during swallowing as the patient makes an effort to achieve an oral seal.

In this study, an obtuse cranial base angle in combination with increased lower anterior facial height may have contributed to the posterior position of the mandible. Bishara et al[7] and Mars and Houston[8] have observed similar findings in their study. Ross[21] in his surgically treated cleft patients also found the mandible to be smaller in size and positioned more posteriorly. Da Silva et al[22] compared mandibular morphology in operated and unoperated CUCLP samples and concluded that there is no statistical difference between the two groups. He further stated that surgical procedure has little influence on the mandibular growth pattern. Although the mandible may not be directly affected by the cleft, it may have an altered growth pattern because of the affected maxilla and functional factors.

Face height measurements in the unoperated cleft patients showed an excess vertical growth tendency. The mandibular plane angle and lower anterior facial height were increased when compared with normal controls, whereas the facial axis, upper facial height to lower facial height ratio, and Jaraback measurements were decreased indicating a more vertical growth direction. Ortiz-Monasterio et al[11] and Bishara et al[12] observed similar findings. The increase in mandibular plane angle may be because fo increased anterior maxillary growth or decreased posterior maxillary growth but may also reflect the intrinsic mandibular growth pattern.[23]

Further clinical research must also be conducted to identify treatment protocols that will interfere least with the normal growth and development of the maxilla in cleft lip and palate patients and permit normal growth to proceed.

CONCLUSIONS

- A normal growth potential exists for the anteroposterior development of the maxilla in unoperated cleft lip and palate patients.
- Because lateral cephalometric studies only examine

the midsagittal structure, 3-D CT studies of the unoperated cleft are necessary to further reveal the parasagittal morphology of the maxilla on the cleft and the noncleft side.

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