Isolated Influences of Surgery Repair on Maxillofacial Growth in Complete Unilateral Cleft Lip and Palate

Zhi-Wei Zheng, MM,* Yi-Ming Fang, PhD,† and Chong-Xiang Lin, MM‡

Purpose: The purpose of this study was to investigate the maxillofacial morphologic characteristics of children with complete unilateral cleft lip and palate (UCLP) with and without surgical correction and better understand the relation between surgery and inhibition of maxillary growth.

Material and Methods: Lateral cephalometric radiography was performed in 3 groups of 9-year-old children: 1) 20 whose UCLP was repaired in infancy; 2) 20 who had no surgical repair or any relevant treatments; and 3) 20 without UCLP who served as controls.

Results: Marked morphologic deviations of patients in groups 1 and 2 compared with the control group were observed in the cranial base angle, maxillary heights, length and shape of the mandible, and anteroposterior jaw relation. Facial morphology was similar between groups 1 and 2 except for less vertical maxillary height and more obtuse gonial angle in group 1.

Conclusion: The facial morphology of children with UCLP differs markedly from that of normal children. The differences can be ascribed to the difference in the primary anomaly in the UCLP groups, but isolated surgery has minor effects on growth disturbances.

Maxillary retraction is a well-recognized phenomenon in a proportion of patients with surgically corrected complete unilateral cleft lip and palate (UCLP).1-4 Two hypotheses have been proposed to explain the tendency toward retraction in patients with corrected UCLP: an intrinsic hypothesis suggests an inherent tissue deficiency and an iatrogenic hypothesis suggests a surgical influence. Developmental tissue deficiency accounts for hypoplasia of the maxilla, and surgical repair is considered detrimental to the growth of the maxillary skeleton by devascularization, disturbance of the periosteum, or the restrictive effect of the scar. Understanding the primary etiologic factors resulting in abnormal maxillary growth enables proper treatment planning, minimization of orthodontic treatment time, and fewer major secondary corrective surgeries.5

In contrast, the separate effects of surgery on craniofacial growth in individuals with UCLP are difficult to assess clinically and remain controversial. This could be a consequence of the heterogeneity of samples. The main problem is that cleft repairs in developed countries are routinely performed in infancy. Previous comparative studies using controls without cleft have confounded the inference of the effects of surgery, because intrinsic deficiency secondary to the cleft affects postoperative maxillary growth.2,6 However, there are people in developing countries and remote areas who have had no early surgery because of lack of facilities, lack of awareness, or socioeconomic circumstances.
Only a few studies have described the isolated influences of surgery on cephalometric features of patients with UCLP. Unfortunately, some findings are inconclusive because of their methodologic limitations. Therefore, the purpose of this study was to identify the influences of surgery on craniofacial morphology by evaluating the growth of patients with UCLP with and without surgical correction.

**Materials and Methods**

**PATIENTS**

In this retrospective comparative study, facial morphology was examined by cephalometric radiographs in prepubescent Chinese children with nonsyndromic complete UCLP with absence of a soft tissue bridge (Simonart’s band). Because of the small sample of patients with surgically untreated UCLP, the sample size was established at 20 when 20 acceptable sets of slides were available for each group. Because of the retrospective nature of the present study, it was granted an exemption in writing by the institutional review board.

Surgically treated participants included 20 consecutive children 9 years of age (range, 8.8 to 9.9 yr) before the bone grafting procedure (group 1). All patients were operated on from 2000 through 2002 by 1 experienced surgeon (Y.-M.F.) and were treated exclusively according to the surgical protocol of The First Affiliated Hospital of Wenzhou Medical University (Wenzhou, China; Table 1).

Briefly, presurgical orthopedic treatment (ie, infant plate) was performed from birth to 4 months of age. Lip closure according to a modified rotation-and-advancement technique and 1-stage palate repair using a 2-flap technique were performed separately at 4 to 8 and at 12 to 14 months of age, respectively. They had no previous orthodontics, protraction headgear, alveolar bone grafting, secondary nasolabial surgery, velopharyngeal surgery, or distraction osteogenesis.

Untreated participants consisted of 20 children with UCLP 9 years of age (range, 8.4 to 10.8 yr) without cleft repairs or any relevant treatments (group 2). The only difference between the 2 UCLP groups in this study was surgical treatment versus no treatment of the lip and palate. Twenty age- and gender-matched healthy students of Han nationality with normal profile served as controls (group 3). None of them had ever undergone any orthodontic treatment or had any trauma of the facial soft tissue. A comparison of the 3 groups is presented in Table 2.

**CEPHALOMETRY**

A lateral cephalogram was obtained for each patient in a standardized manner, with the head in natural position and with the teeth in centric occlusion. The settings of thecephalostat machines and the magnification of the cephalograms (enlargement, 10%) were the same for all patients. All measurements were traced and measured by the same researcher (Z.-W.Z.) without knowledge of the subjects’ treatment histories. Figure 1 illustrates the landmarks and reference planes used in this study. For linear measurements, the enlargement (10%) was corrected. Intra-investigator reliability was assessed by tracing 15 randomly selected cephalometric radiographs at 2-week intervals. Measurement errors (intra-examiner reliability) were assessed by error statistics, which averaged 0.5 mm for linear variables and 0.6 for angular variables.

**STATISTICAL ANALYSIS**

Generalized linear models using SPSS 17.0 (SPSS, Chicago, IL) were performed to assess the adjusted means for craniofacial measurements among groups after adjusting for gender, age, and cranial base size, when indicated. When relevant overall differences among the 3 groups were found, comparisons among groups were undertaken. Probabilities less than .05 were accepted as significant.

**Results**

Cephalometric parameter comparison analysis determining cranial morphology showed marked differences between patients with UCLP (groups 1 and 2) and controls (group 3), whereas minor differences were found between patients with (group 1) and without (group 2) surgical treatment (Table 3). Figure 2 shows the mean profilograms of the 3 groups.
COMPARISON OF CRANIOFACIAL MORPHOLOGY BETWEEN PATIENTS WITH UCLP AND CONTROLS

Anterior overjet ($P < .001$) and anteroposterior jaw relation at the alveolar (angle formed by the A point, nasion, and B point [ANB], $P < .01$) and basal (angle formed by the anterior nasal spine, nasion, and pogonion [ANS-N-Pog], $P < .05$) levels in groups 1 and 2 (with and without surgical repair, respectively) differed substantially from healthy cephalometric standards (Fig 3).

Compared with the controls, the cranial base angle (angle formed by the nasion, sella, and basion [N-S-Ba], $P < .05$) was significantly increased in groups 1 and 2 and the depth of the bone pharynx (basion to posterior maxillary point; $P = .002$) was significantly increased in group 2. The ramus length (gonion to gnathion [Go-Gn]) and total length (articulare to gnathion [Ar-Gn]) of the mandible was increased in groups 1 and 2, but only patients in group 2 showed significant differences ($Go-Gn, P = .002; Ar-Gn, P < .001$).

In group 1, the anterior and posterior heights of the basal maxilla (N-ANS and S-PMP, respectively) were shorter and the gonial angle was more obtuse compared with the controls, which was significant for the gonial angle ($P < .05$) and highly significant for the S-PMP ($P = .002$). Group 2 showed the opposite trend, with increased N-ANS and S-PMP and smaller gonial angle, but only the N-ANS ($P = .02$) showed significant changes.

COMPARISON OF CRANIOFACIAL MORPHOLOGY BETWEEN PATIENTS WITH SURGICALLY TREATED AND UNTREATED UCLP

Results of the generalized linear models showed that compared with group 2, the radiographs in group 1 had significantly decreased anterior and posterior maxillary heights (N-ANS, $P = .004$; S-PMP, $P < .001$), a shorter total mandible (Ar-Gn, $P = .022$), and a more obtuse gonial angle ($P = .01$). The relation between the jaws at the alveolar and basal levels (ANB, $P > .05$; ANS-N-Pog, $P > .05$) was similar in groups 1 and 2, although significant differences were found compared with controls. No meaningful differences were found for the other craniofacial data.

**Discussion**

**STUDY DESIGN AND METHODOLOGY**

Although many authorities throughout the world accept that surgical cleft repair is responsible for midfacial growth disturbances in UCLP, disagreement...
remains as to which observed deviations are part of the primary anomaly and which are caused by surgical interventions.\textsuperscript{1,2,8,11,12} The main problem is the lack of available patients without surgical treatment. Therefore, the observed effect on craniofacial growth is generally attributed to the combined effects of the cleft (intrinsic and functional factors) and surgical repair. In this study, the 2-way comparison of results after completion of repairs with the untreated and control groups provided another important alternative for understanding the multiple perspectives of the isolated influence of surgical treatment on the craniofacial morphology if the effects of the cleft and repair are cumulative when combined.

Although there are many patients with untreated UCLP in developing countries, it is especially important to adjust for differences in baseline character-

<table>
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<th>Group 1—Operated</th>
<th>Group 2—Unoperated</th>
<th>Group 3—Normal</th>
<th>( P ) Value*</th>
<th>Group 1 vs 2</th>
<th>Group 1 vs 3</th>
<th>Group 2 vs 3</th>
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<td>.138</td>
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Abbreviations: A, A point; ANS, anterior nasal spine; Ar, articulare; B, B point; Ba, basion; Gn, gnathion; Go, gonion; gonial angle, inclination of the mandibular plane in relation to the tangent to the posterior border of mandibular ramus through articulare; li, incisal edge inferior; Is, incisal edge superior; Me, menton; MP, mandibular plane (tangent to lower border of the mandible through the menton); N, nasion; PMF, posterior maxillary point (construct created by dropping a perpendicular to the line from the anterior nasal spine passing through the posterior hard palate from the pterygomaxillary fissure); Pog, pogonion; PP, palatal plane; S, sella.

* Overall differences among the 3 groups using generalized linear models. Comparisons between groups were undertaken only when significant overall differences were found.

\textsuperscript{y} Adjusted for cranial base size (S-N).


Marked morphologic deviations of patients with UCLP (groups 1 and 2) compared with control
subjects (group 3) were observed for the cranial base angle, maxillary heights, length and shape of the mandible, and anteroposterior jaw relation. In contrast, compared with patients with untreated UCLP, there was no greater restraint of maxillofacial growth after surgery, except for shorter vertical maxillary height and more obtuse gonial angle. In other words, although a difference between the cephalometric morphologic characteristic of the cleft face and normal face did exist, such differences were not necessarily the result of surgery, but could have been due to intrinsic effects.

CONSEQUENCES OF SURGERY FOR MAXILLARY DEVELOPMENT

The present study found that the isolated influences of surgery on craniofacial growth were principally on the vertical development of the basal maxilla, anteriorly and posteriorly, as evidenced by the shorter anterior and posterior heights of the basal maxilla (N-ANS and S-PMP, respectively) in patients with surgically corrected UCLP. This view is supported by previous studies that have reported decreased maxillary height after surgical treatment of the lip and palate and only the lip. Liao and Mars attributed the deviation of the N-ANS in the surgically untreated group to functional effects, and previous studies have indicated the deviation can be partly corrected by lip and palate repair, as evidenced by the N-ANS differing significantly from the corresponding untreated N-ANS (P = .004) but within normal limits (P = .48). The decrease of the S-PMP was similar to that of the N-ANS in the surgically treated group; accordingly, the inclination of the palatal plane (S-N-PP) was the same for the surgically treated patients and controls.

The present study also showed that the isolated influences of surgery on craniofacial growth were minor on the anteroposterior developmental dimensions of the basal maxilla, as evidenced by the similar depth of the bony pharynx (Ba-PMP), total length of the basal maxilla (PMP-ANS) and alveolar maxilla (PMP-A), and protrusion of the maxilla (SNA). In contrast to previous observations, although the length and protrusion of maxilla were decreased in the surgically treated group, these differences failed to reach statistical relevance in any group. Consistent with this observation, Mars and Houston and Chen et al found that compared with normal controls, the SNA was slightly but not meaningfully smaller in patients with surgically treated and untreated cleft. Previous studies also have found favorable anteroposterior maxillary growth in patients with unoperated UCLP. Taken together, the decreased basal maxillary height (N-ANS, S-PMP), similar length and protrusion of the maxilla, and similar depth of the bony pharynx support the hypothesis that growth disturbances of the maxilla are restricted to the size of the basal maxilla, which are major in height and minor in length.

MANDIBULAR GROWTH

Another effect of surgery was on the growth pattern of the mandible. Patients in the surgically treated group had a more obtuse mandibular angulation (gonial angle) than did controls, but no differences in mandibular length (Ar-Go, Go-Gn, Ar-Go), or protrusion (SNB) were found between patients with surgically treated UCLP and control subjects. This confirmed results obtained in other studies. Because these findings were present in older patients rather than newborns, Liao and Mars indicated that the changes in mandibular position and shape were not related to an intrinsic or iatrogenic effect but to compensatory effects secondary to...
decreased patency of the nasopharyngeal airway in patients with UCLP.

**ANTEROPOSTERIOR JAW RELATION**

The tendency toward a less protruded alveolar maxilla (SNA) and a more protruded alveolar mandible (SNB) in patients with UCLP (with or without surgery) was responsible for the low anteroposterior jaw relation at the alveolar level (ANB). This finding supports the hypothesis of intrinsic developmental deficiency in the maxilla, with the deficiency occurring mostly in the anterior part. However, in contrast to observations of more severe growth disturbances after cleft closure, no meaningful differences in anteroposterior jaw relation at the alveolar (ANB) and basal (ANS-N-Pog) levels were observed between the surgically treated and untreated groups. This corresponds to the results described by Bishara and Yoshida et al who found that maxillary retrusion in UCLP is not caused by surgery but by an intrinsic deficiency. Moreover, a similar trend was found for anterior overjet.

**CRANIAL BASE GROWTH**

Compared with the control group, the cranial base angle was increased in the UCLP groups. This confirms results obtained in other studies. However, disagreement remains as to whether such differences...
are practically relevant, because it seems unlikely that cleft repair could affect the shape or size of the cranial base in patients with UCLP. This finding also could explain the changes in Ba-PMP, by horizontal displacement of the Ba, in the present patients.

CLINICAL RELEVANCE

The finding that the effects of isolated surgical repair on growth disturbances are minor in patients with UCLP has clinical implications. Palate repair has been considered very harmful to maxillary growth; therefore, some surgeons have advocated later closure of the hard palate with a 2-stage procedure in response to the hypothesis that major disturbances in maxillary growth are attributable to palatal surgery. The main disadvantage is the high incidence of defective inferior articulation. Importantly, the results of this study indicate that it is not the operation but the cleft per se that impairs craniofacial growth; therefore, early closure of the hard palate could be of greater therapeutic value to achieve normal development of speech and psychology. This view is further supported by previous studies that have reported that delayed palate repair has no benefit on craniofacial growth. It also reconfirms that promoting maxillary growth during the growth period is extremely important.
LIMITATIONS OF THIS STUDY

The present study has certain limitations. First, the sample is small. Cleft repair is routinely performed in infancy, which eliminates the opportunity to observe and study a large number of patients with unrepaired clefts at a later stage of development. Nevertheless, it is a large growth outcome study performed on patients with surgically treated and untreated patients with UCLP. Second, this retrospective study has problems common to all retrospective studies, and large percentages of patients did not have documentation of cleft size because infant maxillary dental casts were not available. Therefore, cleft size and cast analysis was not compared for the 3 populations, which will be incorporated into another study. However, there was no bias in allocating the patients. That is, consecutive patients with various cleft sizes were treated. Third, the study had no available group on which to test the isolated effects of cheiloplasty and palatoplasty, because these procedures were performed during the first 2 years of life. However, the results would be similar because even cumulative maxillofacial disturbances attributable to these procedures were minor at 9 years of age in this study. However, postpubertal growth will need to be followed in another study. Fourth, the lateral cephalogram is a 2-dimensional representation of a 3-dimensional structure, and only sagittal and vertical relations were evaluated.

In this study, meaningful morphologic deviations of patients with UCLP from normal control subjects were
observed in the study. However, only a decreased basal maxillary height and a more obtuse gonial angle were found compared with the untreated group. Contrary to expectations, anteroposterior development of the maxilla was not affected by the surgical repair by 9 years of age. Overall, cumulative maxillofacial growth disturbances attributable to congenital cleft and iatrogenic surgery were not substantially worse than those attributable to cleft alone in patients with UCLP, suggesting that isolated surgery repair had minor effects on growth disturbances. Future studies with a large sample and more long-term follow-up could confirm the generalization of the results.

References