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Cephalometric analysis of craniofacial morphology and growth in unrepaired isolated cleft palate patients

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ABSTRACT

Objective: The aim of this study is to analyze the craniofacial morphology in patients with unrepaired isolated cleft palate (UICP) at childhood, adolescence and adulthood, in order to assess the influence of nonsurgical factors on the craniofacial growth in these patients.
Material and methods: Lateral and posteroanterior cephalograms of 106 non-syndromic UICP patients and 102 normal matched controls were obtained and analyzed. Patients and controls were divided into three subgroups: children (5–7 years), adolescents (12–14 years), and adults (>18 years).
Results: UICP patients in childhood showed a shortened cranial basal length; reduced bony nasopharyngeal height; short maxillary depth and height with a posterior positioned maxilla and an increased width of the nasal cavity, maxilla and orbit; and a shortened mandibular length and height. UICP patients in adulthood showed a normal nasopharyngeal and mandibular morphology. However, the patients in this subgroup still showed a shortened cranial basal length, and short maxillary depth and anterior height with increased width of the nasal cavity, maxilla and orbit.
Conclusions: Craniofacial morphology and growth in patients with UICP were significantly affected by nonsurgical factors. Growth of the cranial base and upper face were absolutely reduced, while growth of the bony nasopharynx and mandible were only postponed.

1. Introduction

Restricted development of craniofacial structures is often seen in patients with cleft palate. The causes of this limitation of growth are still controversial. Some previous reports (Mars and Houston, 1990; Mazaheri et al., 1967; Ross and Coupe, 1965) attributed it to previous surgical repairs. However, others (Bishara, 1973; Dahl, 1970; Smahel, 1984) introduced factors aside from surgical manipulation (induced traits) that might play a role, such as: (1) genetic pattern (inherited trait), and (2) adaptive changes resulting from the mechanical presence of the cleft (acquired traits). In order to analyze the adverse effects of these nonsurgical factors on craniofacial growth and morphology, all variables introduced by surgery should be eliminated. In that case, the unrepaired cleft patient is the most appropriate subject to be investigated.

In the past, many cephalometric studies have been performed on craniofacial morphology in infants with unrepaired isolated cleft palate (UICP). Bishara (1973); Dahl et al. (1982) and Hermann et al. (2002) all found that infants with UICP showed a short maxilla, a reduced posterior maxillary height, an increased posterior maxillary width and nasal width, a short mandible, a reduced posterior height of the mandible, and a reduced pharyngeal depth and height.

However, relatively few studies have reported on adult patients with UICP. Also, these few studies yielded inconsistent results. Yoshida et al. (1992) found that UICP patients with a mixed dentition had an almost normal craniofacial morphology, but patients with a permanent dentition showed a retruded maxilla, a short hard palate, an inferior rotation of the mandible and a relative maxillary retrusion. This is in contrast with the results of Atherton (1967), concluding that some deformities, which were obvious in

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young patients, were almost undetectable in adult patients. Although the results are not consistent, the aberrant findings in both reports do suggest a potential adverse effect of the nonsurgical factors on craniofacial morphology. However, the question remains, if there is a continuing intrinsic defect and craniofacial growth is absolutely reduced, or if the influence is only temporary and craniofacial growth is only postponed.

Fortunately, most patients with a cleft palate now undergo surgery early in life (Bishara et al., 1986), and the number of individuals in late childhood or adulthood who have not had surgical correction is rapidly diminishing in most areas of the world. On the other hand, in order to accurately assess the influence of the nonsurgical factors on craniofacial morphology and growth, not only infants with UICP should be analyzed. Their observations should be correlated with results of patients with UICP from older age groups. Only in developing countries and remote areas where early surgery is not readily available, there is still an opportunity to examine a limited number of patients with unoperated cleft palates at later stages of development. With the current lack of knowledge in mind, study of these UICP patients may help us to understand the influence of nonsurgical factors.

The purpose of this study is to analyze the craniofacial morphology in children, adolescents and adults with UICP, in order to assess the influence of the nonsurgical factors on the craniofacial morphology and growth in these patients and in particular to elucidate whether nonsurgical factors absolutely reduce craniofacial growth or only postpone it.

2. Material and methods

2.1. Subjects

One hundred and six Chinese patients with a non-syndromic unoperated isolated complete cleft of hard and soft palate (UICP) from the southwest of China, who were referred for palatoplasty to the Department of Cleft Lip and Palate, West China College of Stomatology, Sichuan University, served as subjects for this study. Measurements of 102 normal southwestern Chinese, matched for age and gender and with Angle class I occlusion without any remarkable craniofacial deformities, served as controls for this study. Patients were divided into three subgroups: child group ranging from 5 to 7 years, adolescent group ranging from 12 to 14 years, and adult group older than 18 years (Table 1). Informed consent was obtained from all patients and controls following institutional review board approval.

2.2. Methods

Lateral and PA cephalograms of all patients and controls were taken in the same cephalostat. Patients and controls were orientated to the Frankfurt horizontal plane with teeth in occlusion. The digital radiographs were analyzed directly with WenCeph 7.0 (Rise Corporation, Sendai, Japan).

Table 1
Distribution of patients and controls according to age and sex.

Sex	UICP		Controls	
Age (years)	Male	Female	Male	Female
5–7	12	24	11	21
12–14	14	26	14	26
>18	9	21	10	20

UICP: unrepaired isolated cleft palate.

The following landmarks were identified on each lateral cephalogram: Ba, basion; S, sella; N, nasion; ANS, anterior nasal spine; PNS, posterior nasal spine in controls; Pl, palatale (the most posterior point of the palatal processes in UICP); Ptm, pterygomaxillary fissure; Pmp, pterygomaxillarepalatinum (the intersection of the palate plane with the pterygomaxillary fissure in UICP); Cd, condylion; Go, gonion; Gn, gnathion; Pgn, prognathion; Pg, pogonion; Ii, incisiorinferius; A, point A; B, point B (Fig. 1A) (Smahel, 1984). On each PA cephalogram, the following landmarks were identified: Lo, lateroorbitale (the intersection between the lateral margin of the orbit and linea innominata); Apt, apertion (most lateral point of the nasal cavity); Mo, medioorbitale (most medial point of the orbital orifice); Mx, ectomaxillare (intersection of lateral contour of upper alveolar process and lower contour of maxillozygomatic process of maxilla); Zyg, zygion (most lateral points on the zygomatic arch) (Fig. 1B) (Motohashi et al., 1994).

From these landmarks, various linear and angular measurements were derived. The parameters used in this study were as follows:

1 Cranial base: Anterior length (S–N), Posterior length (S–Ba), Total length (N–Ba), Cranial base angle (NSBa)
2 Bony nasopharynx: Length in UICP (Pmp-Ba), Height in UICP (Pmp-S); Length in controls (PNS–Ba), Height in controls (PNS–S)
3 Upper face: Maxillary depth in UICP (ANS-Pmp), Maxillary depth in controls (ANS-PNS), Anterior height of the upper face (N-ANS), Posterior height of the upper face in UICP (Pmp-NSL), Posterior height of the upper face in controls (PNS-NSL), Maxillary sagittal position (SNA); Upper facial width (Lo–Lo′), Inter-orbital distance (Mo–Mo′), Mid-facial width (Zyg–Zyg′), Nasal width (Apt–Apt′), Maxillary alveolar width (Mx–Mx′)
4 The lower jaw: Mandibular body length (Gn-Go), Mandibular ramus length (Cd-Go), Total mandibular length (Gn-Cd), Anterior height of the mandible (Ii-Pgn), Mandibular sagittal Position (SNB), Sagittal position of chin (SNPg); The angle of the mandibular plane (SN/GoPgn); The angle between the palatal plane and mandibular plane (ANSPmp/GoPgn in UICP andANSPNS/GoPgn in controls); Bicondylar width (Cd–Cd′), Bigonial width (Go–Go′)
5 Maxillary mandibular relationship: ANB

The same investigator (Yi Xu) identified all landmarks and derived all measurements twice, with an interval of two weeks. The average value of each pair of measurements was used for statistical analysis. Intra-observer concordances of four measurements related to the Pmp (Pmp-Ba, Pmp-S, Pmp-NSL, and ANS-Pmp), which were most likely to be affected by landmark identification, were analyzed at the childhood age subgroup to investigate the landmark identification reliability of this method.

2.3. Statistical analysis

Pearson correlation coefficients (Pearson r) were used to test intra-observer concordance. A Student t-test was used to test the significance of differences between same age subgroups of UICP and controls and also between different age subgroups of UICP or controls. The level of significance was set to 0.05.

3. Results

The Pearson r coefficients of Pmp-Ba, Pmp-S, Pmp-NSL, and ANS-Pmp were 0.996, 0.993, 0.990, and 0.994 for intra-observer measurements suggesting high landmark identification reliability of this method.