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Clinical Paper
Craniofacial Anomalies

Comparison of the adult three-dimensional craniofacial features of patients with unilateral craniofacial microsomia with and without early mandible distraction

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Abstract. This study was conducted to analyze the long-term facial growth of patients with craniofacial microsomia (CFM) after early mandible distraction osteogenesis (DO), and compared adult three-dimensional (3D) craniofacial features of patients with and without early mandibular DO for Pruzansky grade II deformities. The study included 20 patients: 9 with early mandible DO (the DO group) and 11 without previous treatment (the NDO group). Longitudinal radiographs were measured for growth changes after DO. The 3D craniofacial images were constructed to compare the craniofacial forms between the two groups. The patients with early DO presented 8 to 9 mm forward and downward maxillary growth and 4.6 mm limited forward and 17.3 mm substantial downward mandibular growth. The ramus length ratio (affected/nonaffected) was 90.8% at DO completion and decreased to 69.5% at growth completion during 13 years of follow-up. Both groups showed obvious craniofacial asymmetry, as indicated by occlusal plane canting, chin deviation, transverse and vertical condyle positions, and mandibular contours. Although all the bilateral differences were higher in the NDO group than in the DO group, no statistical differences were found. Early mandible distraction could not alter the inherent facial growth pattern in patients with grade II CFM. Limited changes are derived for definitive facial correction with early DO.

Key words: craniofacial macrosomia; mandibular distraction osteogenesis; 3D craniofacial analysis; craniofacial growth and development.

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Craniofacial microsomia (CFM) refers to a broad spectrum of congenital malformations resulting from the variable dysmorphogenesis of craniofacial structures derived mainly from the first and second branchial arches[1]. The severity of mandible deformities is graded according to the Pruzansky classification system and Kaban modification[2,3]. Although the classification scores are inherently inaccurate and variable, they remain the most common medical tool for determining treatment modalities for CFM[4].

Orthodontic and surgical decisions for CFM are influenced by the severity of facial skeletal involvement as well as the functional and psychological needs of the patients, from camouflage orthodontic treatment or functional appliance, to costochondral graft or free flap treatments. In 1992, distraction osteogenesis (DO) was first proposed as an effective and safe technique for expanding mandibular dimensions in young patients with gross facial asymmetry[5]. In our previous study, multiplanar mandible DO proved to be effective for correcting facial skeleton asymmetry and lip canting in young children with CFM. The 1-year cephalometric follow-up revealed relapses in mandibular ramus height of 30% and chin deviation of 1.6 mm[6]. Other studies have demonstrated similar relapse of the mandible ramus after early DO and progression of mandibular asymmetry and occlusal plane canting[7,8]. A comprehensive literature review provided no evidence for the long-term effectiveness of early DO in patients with CFM who were followed up from 1 to 5 years[9]. A three-dimensional (3D) computed tomography (CT) study reported that 50% of patients relapsed 1 year after DO and exhibited progressive deterioration for 3 years when the changes in the mandible volume were measured[10].

A longitudinal panoramic study plotted the growth curve changes in the mandibular ramus height of patients with CFM. The growth rate of patients with grade I/IIa CFM was similar to that of controls, and the growth rate of patients with grade IIb/III CFM gradually decreased compared with that of controls[11]. Longitudinal mandibular vertical growth was observed in patients with CFM who were treated with or without early mandible DO until the completion of facial growth. The ratio of the affected to the nonaffected ramus height returned to the original asymmetry in patients with early mandible DO[12]. Mandible DO does not seem to be a permanent long-term treatment for CFM; it was reported that 90% of patients exhibited the recurrence of facial asymmetry and required orthognathic surgery (OGS) to improve the treatment outcome[13].

To date, few long-term longitudinal studies have investigated facial growth until maturation in CFM patients with mandible DO. The aims of the current study were as follows: 1) to analyze long-term facial growth in adult patients with CFM after early mandible DO; and 2) to compare the adult 3D craniofacial features of patients with and without early mandible DO for Pruzansky grade II deformities.

Materials and Methods

This retrospective cohort study examined 20 consecutive adults with unilateral CFM who received definitive orthodontic/orthognathic treatments between 2012 and 2015. The indications for surgical correction were as follows: patients' concern about obvious facial asymmetry; 2) occlusal and functional demand; and 3) requirements for psychological support. These patients were grouped according to their previous treatment into the DO group, comprising patients with early mandible DO (n = 9), and the NDO group, comprising those without early mandible DO (n = 11).

The inclusion criteria were as follows: 1) unilateral CFM with grade II mandible deformities; 2) complete traceable radiographic records for previous treatment in the DO group; 3) the completion of facial growth and maturation of physical growth; and 4) CBCT record at the time of the completion of facial growth and before definitive facial correction in both groups. Patients with major craniofacial trauma and incomplete records were excluded. For the DO group, 11 patients from our previous study were included[6], and two patients in our previous study were excluded. One patient had temporomandibular joint (TMJ) ankylosis, and the other claimed no further treatment needs and discontinued follow-up.

Longitudinal facial growth in the DO group

The mandible DO was performed at ages ranging from 5 to 9.4 years. Nine patients (3 male and 6 female; 5 and 4 with CFM of the right and left side, respectively) were followed until growth completion, with a mean age of 21.04 (standard deviation [SD] 3.37) years and a mean follow-up of 13.08 (SD 3.25) years. Previous treatment computed tomography (CT) or cone beam computed tomography (CBCT) data were not available for these patients. Therefore, longitudinal facial growth was measured using panoramic and lateral cephalometric radiographs. Facial growth was investigated at the time points of T0 (completion of DO) and T1 (complete growth maturation and before definitive treatment). The ramus length ratio was calculated from panoramic radiographs as the affected/nonaffected ramus length. The reference lines in the lateral cephalometric radiograph were defined as follows: X-axis, 7° below the sella nasion (SN) line; and Y-axis, perpendicular to the X-axis through the sella. The linear distance of the anterior nasal spine (ANS) and gnathion (Gn) relative to the X- and Y-axes and the angle of the mandibular plane relative to the SN were measured (Fig. 1).

Comparison of 3D craniofacial features between DO and NDO groups

The NDO group included 11 patients (4 male and 7 female; 6 and 5 with CFM of the right and left side, respectively) with a mean age of 20.25 (SD 2.73) years. The craniofacial morphology was compared at the timing of growth completion and before definitive surgical-orthodontic correction between the 2 groups in cross-section.

For evaluating 3D craniofacial morphology, CBCT was obtained for analysis. The patient's skull was orientated to the natural head posture and in the upright position. The 3D internal reference planes were as follows: 1) the Frankfort horizontal plane (FH), which connects the bilateral orbitale and the nonaffected porion; 2) the midsagittal plane (Mid), the midfacial plane parallel to the patient's true vertical and passing through the nasion; and 3) the coronal plane (Cor), the plane perpendicular to the FH and midsagittal planes and passing through the basion (Fig. 2). The mandible was segmentalised to evaluate its form and shape. The 3D landmarks were identified to measure the distance relative to the three reference planes. The definitions of the 3D landmarks are provided in Table 1. Thus, the transverse, vertical, and sagittal positions of each 3D landmark were identified and compared. For measurement, a plus sign (+) represented landmarks located on the nonaffected side relative to Mid, downward to the FH, and forward to Cor, and a minus sign (−) represented the landmarks located on the affected side relative to the Mid (Fig. 3).

The mandible was further separated into the ramus and body portions. The plane to

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