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Condylar positions before and after bilateral mandibular distraction osteogenesis in children with Pierre Robin sequence

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Abstract. This study aimed to analyze the changes in mandibular morphology after bilateral mandibular distraction osteogenesis (BMDO) in children with Pierre Robin sequence. The positions of the condyles were analyzed in reconstructed threedimensional craniomaxillofacial images obtained for 18 children before and 8-12 weeks after BMDO. Differences between pre- and postoperative parameters were assessed using paired *t*-tests. After surgery, a significant decrease in superior joint space was detected (P < 0.05), but no change in anterior joint space or posterior joint space was observed. The ratio of the distance between gonia and distance between condylion points (GoL–GoR/CoL–CoR) (P < 0.001) and the distances between the condyle and midsagittal plane (P < 0.001) increased after surgery, while the condylar horizontal angle decreased (P < 0.05). No change in condylar vertical angle was noted. After BMDO, the condyle displayed an outward and upward shift, as well as outward rotation along the proximal segment. The mandible body exhibited forward movement with a more significant opening range. These changes were consistent with the extent of the newly formed bone tissue and the improvement in coordination and appearance of the children's facial structures. The long-term effects of changes in condylar position on the development of the maxillofacial structures needs to be studied further.

Key words: Pierre Robin sequence; condyle; joint space; bilateral mandibular distraction osteogenesis.

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Pierre Robin sequence (PRS) is a rare congenital mandibular abnormality with a reported prevalence ranging between 1/8500 and $1/20,000^1$. PRS is character-

ized by micrognathia and glossoptosis, with or without cleft palate. The deformities may result in airway obstruction in the neonate, often accompanied by cyanosis and retraction of the intercostal space and upper fossa during inspiration. There may be serious clinical consequences for children with multiple PRS malforma-

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2 *Liu et al.*

tions, and the mortality rate is estimated to be up to 26%. Also the movement of the epiglottis may be restricted in children with PRS, resulting in chronic malnutrition¹.

Prone positioning is the first-line noninvasive management of PRS. In the case of persistent airway obstruction, a surgical intervention is required, such as tonguelip adhesion, tracheotomy, or mandibular distraction osteogenesis (MDO)². Over the last two decades, distraction osteogenesis has been tested for the correction of temporomandibular joint (TMJ) deformities in animal models as well as in the clinical setting^{3,4}. Bilateral mandibular distraction osteogenesis (BMDO) improves glossoptosis by extending the mandible, thus clearing the airway. This has become the primary choice for the treatment of patients with severe mandibular retrognathia⁵. It is generally suggested that BMDO is performed at an early stage to improve breathing and nutrition in a timely manner, and therefore to improve quality of life.

In BMDO treatment, truncation and facilitated forward movement of the mandible bone segment may cause a shift in position of the teeth, jaws, skull, and/or facial structures. Changes in mandibular morphology and position may affect the fine anatomical structures and function of the TMJ⁶. Furthermore, the bone composition of infants is more flexible and will adapt during reconstruction of the facial structures. Therefore significant changes in the relative positions of the temporomandibular articulation and mandible may occur during the postoperative period after BMDO treatment.

Most perioperative studies have concentrated on changes in the mandible structure from the pathological perspective, with little attention paid to changes in the intact mandible morphology and position during the developmental stages after BMDO, especially the long-term impact on craniofacial development. In this study, the facial structures of patients before and after BMDO treatment were analyzed using reconstructed three-dimensional (3D) craniofacial computed tomography (CT) images. The mandibular morphology was evaluated with quantitative parameters reflecting the relative positions of the condyles. The potential impact of BMDO on the development of the mandibular structures is discussed.

Methods

Setting and patients

This retrospective study was approved by the Ethics Committee of Guangzhou Women

and Children's Medical Centre. Consecutive patients who underwent BMDO for the treatment of PRS in the Department of Oral and Maxillofacial Surgery of Guangzhou Women and Children's Medical Centre from May 2013 to October 2015 were included.

PRS was diagnosed according to the following criteria: (1) micrognathia and glossoptosis with breathing/swallowing difficulties, defined as the need to maintain a lateral or prone position, or the requirement of assisted mechanical ventilation for respiration, and/or need for tube feeding; (2) reconstructed 3D maxillofacial and laryngeal airway CT images confirming significant tongue falling and upper airway stenosis, with the narrowest position detected between the rear end of the tongue and the hyoid bone.

Patients were included in the study if they fulfilled the necessary eligibility criteria, as outlined below.

Eligibility criteria

The following inclusion criteria were applied: confirmed diagnosis of PRS; no previous surgical treatment such as tongue–lip adhesion; preoperative examination showing a normal bilateral TMJ structure with a normal degree of mouth opening; patient able to tolerate surgery under general anaesthesia; the availability of long-term follow-up records (minimum 2 years).

Exclusion criteria were the presence of any other severe congenital deformity, metabolic disease, chromosomal disorder, or dystonia, or the presence of a breathing or eating disorder of unknown cause not directly related to PRS.

Surgical procedure

All patients underwent BMDO under general anaesthesia. A parallel incision was made about 1.5 cm from the inferior border of the mandible close to the mandibular angle. The osteotomy line was designed according to the preoperative 3D CT findings. A 'walking-stick' shaped osteotomy line passing through the mandibular angle was utilized to avoid the tooth bud and condylar neck. After a rest period of 48-72 h, mandibular distraction was performed at a speed of 1.2 mm/day. The positioning of the distractors is shown in Fig. 1. Distraction was performed until the upper and lower jaws were symmetrically aligned or a slight underbite was achieved. The maximum traction distance was limited to 1.5 cm.

Imaging and analysis of the 3D craniomaxillofacial structures

Three-dimensional maxillofacial CT was performed before and at 8–12 weeks after surgery using a Philips Brilliance CT64 scanner with settings of 120 kV, 130 mA, and 0.9-mm layer thickness. Structure reconstruction was then accomplished using Mimics version 10.0 software (Materialise, NV, Leuven, Belgium) to acquire coronal, sagittal, and axial views.



Fig. 1. Positioning of the distractors in this study.

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