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Three-dimensional quantitative evaluation of midfacial skeletal changes after trans-sutural distraction osteogenesis for midfacial hypoplasia in growing patients with cleft lip and palate

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A B S T R A C T

Purpose: Trans-sutural distraction osteogenesis (TSDO) is an alternative method for the early treatment of midfacial hypoplasia in growing patients with cleft lip and palate (CLP). The purpose of this study was to analyze three-dimensional (3D) midfacial skeletal changes after TSDO and to explore the mechanism in this process.

Material and methods: All patients with nonsyndromic CLP who underwent bone-borne TSDO for midfacial hypoplasia from 2005 to 2014 were reviewed in this retrospective study. 3D morphological and quantitative measurement analyses were performed to evaluate midfacial skeletal changes by superimposition of preoperative and postoperative computed tomographic images.

Results: Twenty-six patients with mean age of 11.5 years met the inclusion criteria. The 3D morphological findings exhibited the most significant suture stress changes at the pterygomaxillary suture area, with obvious bone generation in all patients. The whole midfacial skeleton had progressively increased advancement in a craniocaudal direction along the midface segment, associated with morphological changes in skeleton itself. The 3D quantitative measurement findings showed differential advancement of each landmark at the maxillary alveolar, zygomatic bone, orbital rim, and nasal bone, which was consistent with morphological findings.

Conclusions: TSDO allows rotation advancement of the midfacial skeleton to achieve occlusal correction and facial harmony through the mechanism of both suture remodeling and bone remodeling.

1. Introduction

Midfacial hypoplasia, defined as deficient midfacial skeletal growth, frequently develops in patients with cleft lip and palate (CLP) as a result of iatrogenic factors caused by surgical and nonsurgical interventions (Ross, 1987; Oberoi et al., 2008; Hsieh et al., 2010; Lee et al., 2014) and intrinsic growth deficiency (Wu et al., 2011; Delestan et al., 2014; Doucet et al., 2014). Extensive morphologic studies have demonstrated these skeletal and/or dental deficiencies present in the anteroposterior (AP), vertical, and transverse dimensions in the early stages of pediatric growth (Öztürk and Cura, 1996; Schneiderman et al., 2009; Abuhijleh et al., 2014). Therefore, instead of waiting until skeletal maturity, midfacial hypoplasia have been early treated in growing patients because of psychological, functional, and esthetic concerns (Ho et al., 2006; Scolozzi, 2008).

The early treatment options for midfacial hypoplasia mainly include distraction osteogenesis (DO) with Le Fort osteotomy (Scolozzi, 2008) and trans-sutural distraction osteogenesis (TSDO) (Liu et al., 2005; Tong et al., 2015), which both progressively advance the skeleton in association with the expansion of the

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surrounding soft tissue. TSDO has been proved to have the advantage of achieving the advancement of midfacial skeleton by applying force to the immature circummaxillary sutures while successfully avoiding the risk of Le Fort osteotomy in some way, but there is still lack of recognition of the mechanism of the TSDO in growing CLP patients.

Perhaps the most intuitive aspect of the TSDO process relies on the morphological analysis of skeletal changes. Numerous authors have depicted the simulative suture stress distribution and midfacial skeletal changes through three-dimensional (3D) finite element model analyses of maxillary protraction (Lee and Baek, 2012; Kim et al., 2015). Unfortunately, most of these studies have only allowed favorable speculation about clinical skeletal changes from non-cleft patients with mild to moderate maxillary hypoplasia; therefore, the underlying mechanism of the TSDO in growing CLP patients is still unknown, especially in patients with severe forms. Recently, our center introduced bone-borne TSDO in growing CLP patients with midfacial hypoplasia, which essentially is the traction force enhancement of maxillary protraction with anchorage at the lateral nasal wall (Tong et al., 2015). The results of the initial color-coded map analysis of skeletal changes showed unequal advancement of the whole midfacial skeleton with a blurry boundary exactly matching the Le Fort III fracture line. However, these qualitative results still could not provide complete explanations for the mechanism of the TSDO.

Therefore, the purpose of this study was to conduct a quantitative contrastive analysis of 3D midfacial skeletal changes after TSDO, and to explore the sutures and structures involved in this process, thereby increasing the knowledge of the mechanism of TSDO, which may eventually help to optimize its clinical utility.

2. Material and methods

This retrospective study reviewed all types of CLP patients treated with bone-borne TSDO for midfacial hypoplasia at the Department of Cleft Lip and Palate, Plastic Surgery Hospital, Chinese Academy of Medical Science and Peking Union Medical College, between 2005 and 2014. The study was approved by the institutional review board. Patients were selected for both general demographic information and the following inclusion criteria: growing nonsyndromic CLP patients with midfacial hypoplasia identified by clinical profile evaluation associated with class III malocclusion; treatment with bone-borne TSDO according to the same primary treatment protocol by the same surgeon; and complete preoperative and postoperative computed tomography (CT) scan data. In addition, patients who presented with incomplete or poor-quality data records and major or minor complications during the distraction were excluded from the study.

2.1. Trans-sutural distraction osteogenesis technique

The technique of TSDO had been previously described in detail by Tong et al. Briefly, the distraction system consisted of rigid external distractor (RED, Cibei Medical Treatment Appliance Co., Ningbo, China), and nickel-titanium shape memory alloy spring and bone-borne traction hooks (GEE Co., Beijing, China). Within a certain range of deformation, the spring generated a continuous and stable force of approximately 2.45 N/mm. Intraoperatively, the maxillary vestibular incisions were made and the buccal tissue was reflected to expose the pyriform rim and canine root. A hole was drilled on each side approximately 1 cm outside of the lateral pyriform rim and 5 mm above the apices of the teeth through the lateral nasal wall. Two independent traction hooks were introduced through the hole with the caudal ends extending out from the nostril base. The cranial frame of RED was applied with the frame approximately 20°–30° upward to the Frankfurt horizontal plane and the vertical rod 10–12 cm anterior to the nostril base. The spring was then used to connect the hooks to RED, and the initial distraction direction was adjusted 20°–30° anteroinferiorly.

Without a latency period, the distraction commenced immediately after operation, with the traction force increased gradually at a variable distraction rate of 1–2 mm every 2–3 days, which was decided mainly based on the patient's adaptation and age. When significant advancement of the maxillary was observed, the force would continue to be increased slowly to the maximum and maintained until the required advancement was achieved. The adequate positive overjet with a moderate overcorrection and facial contour were used as the clinical guide to discontinue the distraction. The distraction was then followed by a consolidation period of 1–3 months, with gradually decreased traction force. After removal of the device, for those patients with a significant tendency toward relapse, which meant that the normal occlusal relationship could not be established in time after TSDO, a face mask with elastic traction was used at night to maintain the gain. In addition, the orthodontic therapy could start aligning the dental arch accompanying the distraction determined by the team orthodontist (Fig. 1).

2.2. 3D morphological and quantitative measurement analysis

The skull CT images were obtained preoperatively (T0) and immediately the day after the devices were removed (T1), with the exposure conditions at 120 kV and 240 mA.The slice thickness was set at 0.5 mm. The image matrix size was 512 × 512, and the pixel size was 0.35 mm (Aquilion 64; Toshiba, Tokyo, Japan). The CT data were stored in Digital Imaging and Communications in Medicine file format and analyzed using Mimics 10.01 (Materialise, Leuven, Belgium). To achieve visualized and quantitative contrast analysis of 3D images, the data were preprocessed as follows. Briefly, the resultant slice images were converted to create 3D skeleton models with a threshold technique. Then the T0 and T1 3D skeleton models of each patient were registered on one another using the STL global registration function of the software. The superimposition of T0 and T1 was completed automatically with a minimal point distance filter setting at 1 mm. From these superimposition 3D images, the treatment changes were presented visually and multi-directionally, then cropped and manipulated as superimposition models to allow visualization of the midfacial skeleton and cranial base. To further quantitatively analyze the treatment changes, a new measuring coordinate system was established to standardize the orientation of the craniofacial structures. Given that the T0 and T1 3D skeleton models had been registered into a common Mimics default coordinate system, we could choose the anatomical landmarks of T0 to create a new coordinate system for the measurement of both T0 and T1 3D models. The bony landmarks used for creating 3D reference planes included the following points of T0: sella (S), nasion (N), porion (Po), orbitale (Or), and basion (Ba). The Frankfort horizontal plane (FH plane) was defined as the plane that passed through the bilateral Po and Or on the noncleft side (UCLP) and left side (BCLP). The horizontal reference plane (HR plane) was defined as the plane parallel to the FH plane passing through N. The midsagittal reference plane (MSR plane) was defined as the plane perpendicular to the HR plane passing through Ba and S. The coronal reference plane (CR plane) was defined as the plane perpendicular to the HR and MSR plane passing through S (Fig. 2). The midfacial skeletal changes were evaluated through the anatomical landmarks on 3D models, which were first plotted on the surface of the 3D model and their positions then calibrated in the multiplanar reconstruction views. The anatomical landmarks for 3D measurements are described and summarized in Table 1. After that