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Mandibular volumetric increase following distraction osteogenesis☆

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Summary *Introduction:* Mandibular distraction osteogenesis (MDO) for the treatment of Pierre Robin sequence (PRS) enables mandibular lengthening and improves airway and feeding function. It remains unknown how the post-distracted mandibular volume compares to a normal control population. The aim of this study was to analyze mandibular volume and symmetry following bilateral MDO and compare post-distraction measurements to a non-distracted, normal age- and sex-matched control cohort.
Methods: Demographic information and three dimensional-computed tomographic (CT) images were obtained from normal control and distracted PRS patients. Mandibular volume and symmetry indices were calculated and results statistically analyzed. *P* values $\leq$0.05 were considered statistically significant.
Results: 24 CT scans and 48 hemimandibles were analyzed (8 control patients: mean age = 5.6 months, 3 females; 8 distracted patients: mean age pre-distraction = 1.8 months, mean age post-distraction = 5.3 months, 3 females). No complications were encountered in the distracted group. The mean pre- and post-distraction volume in the MDO group measured 7238.1 mm^3 and 15,360.6 mm^3, respectively ($P = 0.0003$) and the mean percent increase in mandibular volume following distraction was 113.3%. The mean symmetry index increased after distraction from 0.91 to 0.95 ($P = 0.31$). Matched normal control mandibles measured 13,488.6 mm^3 versus post-distraction mandibles at 15,360.6 mm^3 ($P = 0.40$). Normal control and post-distraction symmetry indices were 0.99 and 0.95, respectively ($P = 0.68$).

☆ Product used in this study: Materialise Surgicase CMF Pro (Leuven, Belgium) and mandibular distraction devices (Synthes CMF; West Chester, PA).

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Conclusion: Distraction resulted in a significantly increased mandibular volume and an observed preservation in mandibular symmetry. Post-distraction volume was increased compared to normal controls but remained less symmetrical.

Introduction

Pierre Robin sequence (PRS), classically characterized as the triad of microretrognathia, glossoptosis, and airway obstruction (with or without cleft palate),[1,2] is a congenital abnormality with a reported occurrence as high as 1 in 8500 live births.[3] The condition may be associated with a syndromic or nonsyndromic component, with common syndromes including hemifacial microsomia, Treacher Collins syndrome, and, most commonly, Stickler's and chromosome 22q11 deletion syndromes.[4] Complications associated with PRS include severe upper airway obstruction that may result in life-threatening feeding, respiratory, and cardiac complications. Treatment approaches vary and depend on condition severity. Nonsurgical therapy is the first line approach for less severe disease where respiratory function is not compromised and entails prone positioning and/or placement of a secured nasopharyngeal airway. In cases where nonsurgical therapy has failed or where airway obstruction is life-threatening, surgical management is indicated and includes procedures such as glossopexy, tongue-lip adhesion, and mandibular distraction osteogenesis (MDO), with tracheostomy reserved for the most severe of cases (such as those with multi-level airway obstruction).

Since its application for mandibular lengthening,[5–7] MDO has become a highly utilized surgical technique for the treatment of microretrognathia and other mandibular dysmorphologies.[8,9] MDO enables mandibular lengthening and alleviates glossoptosis to ameliorate airway obstruction .[5,8–15] Several studies have attempted quantitative analysis of the effect of MDO on the mandible and surrounding tissues.[11,13,16–20]

To date, it remains unknown how total mandibular volume and symmetry following MDO compares to a normal control population. Thus the aim of this study is to quantitatively both describe the mandibular volume and symmetry before and after bilateral MDO in infants with PRS and compare these measurements to an age- and sex-matched normal control group.

Methods

This is a retrospective study approved by the Yale University Institutional Review Board (HIC: 1101007932) including a series of consecutive patients under the age of one year who underwent bilateral sagittal internal distraction of the mandible. Computed tomography (CT) scans of normal and distracted patients were analyzed using a 3D software platform (Materialise; Leuven, Belgium). Demographic information from all patients included in this study was gleaned from medical charts. Older children and patients without post-operative CT scans were excluded. The normal control group consisted of a series of infants without existing craniofacial disorders or conditions. Mandibular and hemimandibular volumes were obtained from 3D-rendered images of all patients within the pre-distraction and post-distraction groups (Figure 1) and control groups via digital segmentation as described previously.[21–23] Mean mandibular volume was expressed in cubic millimeters (mm^3) ± the standard deviation and mandibular symmetry was calculated as the ratio of the smaller hemimandible over the larger hemimandible. Statistical analysis was performed using Microsoft Excel (Version 14.0.0, Microsoft Office 2011, Microsoft; Redmond, WA) and SPSS Statistics (Version 19, IBM; Armonk, NY). Comparisons between pre- and post-distracted mandibles and symmetry were conducted using a paired *t* test and a two-tailed student's *t* test was used to compare the post-distracted mandible to the normal control mandible and for symmetry comparisons. An observed *P* value of 0.05 or less was considered statistically significant.

Results

For mandibular distraction, pre-operative CT scans were used to assess mandibular morphology and plan mandibular osteotomies and device placement location, if deemed a distraction candidate. In all cases, a linear internal mandibular distractor device (1.0 mm footplate) was used (Synthes CMF, West Chester, PA). After a latency phase of 24 h, the distractors (barrel size range: 15—25 mm) were activated at 2—2.5 mm per day bilaterally until the desired position was reached (a slight class 3 malocclusion or anterior crossbite). The extension arms were removed and a consolidation phase of approximately eight weeks was initiated to facilitate callus ossification and stabilization of the distraction gap. Distraction devices were removed under general anesthesia and no additional osteosynthesis was necessary for distal segment stabilization (Figure 2).

Twenty-four CT scans (48 hemimandibles) were analyzed. Eight distracted patients and eight age- and sex-matched control patients were analyzed (Table 1). Within the mandibular distraction group, three were female and five were male with a mean pre-distraction age of 1.8 ± 2.1 months and post-distraction age of 5.3 ± 3.9 months at the time of pre- and post-operative CT scans, respectively. Indication for distraction included nonsyndromic retrognathia with airway obstruction (6/8 patients), Goldenhar syndrome (1/8), and Stickler syndrome (1/8). Four patients presented with a cleft palate. The mean post-distraction follow-up period was 12.3 ± 6.2 months. The mean time