

Clinical Paper Craniofacial Anomalies

Precise osteotomies for mandibular distraction in infants with Robin sequence using virtual surgical planning

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Abstract. Mandibular distraction osteogenesis (MDO) has become the first-line operation in many centers for the management of obstructive sleep apnea (OSA) in infants with (Pierre) Robin sequence (RS) not relieved by non-surgical approaches. Preoperative virtual surgical planning (VSP) may improve precision and decrease complications for this operation. This article reports a retrospective study of RS infants who underwent MDO for OSA using preoperative VSP and three-dimensionally printed cutting guides performed by one surgeon. Seventeen subjects who had MDO at a mean age of 87 ± 96 days were included. Maxillofacial computed tomography scans were obtained 15 ± 7 days prior to MDO. Osteotomy designs included linear ($n = 4$, 23.5%), inverted-L ($n = 11$, 64.7%), and multi-angular ($n = 2$, 11.8%). Cutting guides were used successfully and osteotomies were created as planned in all cases. Devices were removed 67 ± 15.6 days after placement. Bone formation in the distraction gap was seen in all cases at device removal. All patients had successful airway outcomes. There were no major and four minor complications during the follow-up period of 458 ± 267 days. In conclusion, MDO is a successful procedure for the management of OSA associated with RS in infants, and VSP facilitates its precise design and execution.

Key words: Robin; Pierre Robin sequence; mandibular distraction; virtual surgical planning.

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Mandibular distraction osteogenesis (MDO) has become the first-line operation in many centers for infants with airway obstruction associated with (Pierre) Robin sequence (RS) who have failed non-invasive management¹. This procedure boasts high rates for successful resolution of obstructive sleep apnea (OSA) and avoid-

ance of tracheostomy^{2–12}. Long-term complications associated with the position of the osteotomies, such as injury to developing teeth and the inferior alveolar nerve, however, are common^{1,13–15}.

Preoperative virtual surgical planning (VSP) and the fabrication of three-dimensionally (3D) printed splints and/or cutting

guides for intraoperative use have been shown to improve outcomes and decrease operative times and costs in other areas of maxillofacial surgery^{16–19}. For neonatal MDO, VSP has been reported to aid in device selection and placement, facilitate vector management, and simplify the operation²⁰. Planning may also improve

precision and decrease complications. However, only single case reports of VSP and 3D printing for MDO in infants with RS currently exist in the literature.

The purpose of this study was to present a series of infants with RS who underwent MDO using preoperative VSP and 3D printed cutting guides. The specific aims were to (1) document the protocol used at Boston Children's Hospital for planning and execution of MDO in infants with RS using VSP, (2) evaluate osteotomy designs used to minimize damage to vital structures, and (3) present intraoperative findings and airway outcomes.

Materials and methods

Study design and subjects

This study analyzed a retrospective case series of infants at Boston Children's Hospital with OSA associated with RS who presented from February 2014 to December 2016. Inclusion criteria were the following: (1) a clinical diagnosis of RS made by a member of the Craniofacial Center, including micrognathia, glossop-tosis, and airway obstruction; (2) OSA that could not be completely treated by non-operative approaches and/or intubation with inability to extubate; (3) use of MDO by one surgeon (C.M.R.); (4) use of preoperative 3D virtual surgical planning and 3D printing of cutting guides. Subjects were excluded if they did not have pre-surgical VSP or if the virtual plan was not utilized in the operating room. This study was approved by the Institutional Review Board of the Center for Applied Clinical Investigation at Boston Children's Hospital.

Variables

Study variables included gestational age, birth weight, sex, syndromic diagnosis, age at presentation to Boston Children's Hospital, age at MDO, dates of preoperative maxillofacial computed tomography scan (CT), use of intubation for preoperative CT, need for repeat CT due to motion artifact, dates of pre- and postoperative polysomnograms (PSGs), dates of VSP Web session, intraoperative findings, overall severity score of OSA from the PSG as scored by the reading sleep medicine physician based on a compilation of all measured parameters (none = 0, minor = 1, minor-moderate = 2, moderate = 3, moderate-severe = 4, severe = 5)²¹, type of osteotomy used for MDO, need for additional airway inter-

vention after MDO, and surgical complications during the follow-up period.

Perioperative management

All patients were managed pre- and post-operatively in the neonatal intensive care unit. Prior to MDO, all non-intubated patients failed non-operative airway management including positioning maneuvers, insertion of a nasopharyngeal tube, and/or use of continuous positive airway pressure therapy. Direct laryngoscopy and bronchoscopy were performed prior to MDO in all non-intubated patients to rule out laryngomalacia, tracheomalacia, and subglottic stenosis as contributors to airway obstruction. Non-intubated patients had a pre-MDO PSG to confirm and characterize the OSA and a postoperative PSG at the end of active distraction prior to the removal of the activation arms. All PSGs were performed at the Boston Children's Hospital Sleep Center and were read by a pediatric sleep medicine physician.

Virtual surgical planning

The maxillofacial CT for each patient was uploaded to a third-party vendor for virtual surgical planning (3D Systems, Inc., Gold-

en, CO, USA) and a Web session was scheduled between the surgeon and a biomedical engineer using GoToMeeting (Citrix, Inc., Santa Clara, CA, USA). Prior to the Web session, the biomedical engineer segmented the CT volumes into relevant anatomical units to highlight structures such as developing teeth and the inferior alveolar nerve and to facilitate virtual surgical movement.

During the Web session, bilateral virtual osteotomies were drawn by the biomedical engineer based on input from the surgeon. These osteotomy positions and configurations were customized for each patient to: (1) minimize or avoid damage to developing dental structures, (2) avoid binding of the mobilized distal segment against the proximal segment during distraction, (3) avoid bringing the coronoid process forward with the distal segment, as this could lead to impingement with the zygomatic arch during distraction, (4) provide sufficient bone in each segment for device fixation, (5) achieve the desired vector for distraction, and (6) match the distraction vector between sides²². Common osteotomy designs included linear oblique, inverted-L, and multi-angular (Fig. 1). After completion of the virtual osteotomies, a digital version of the device

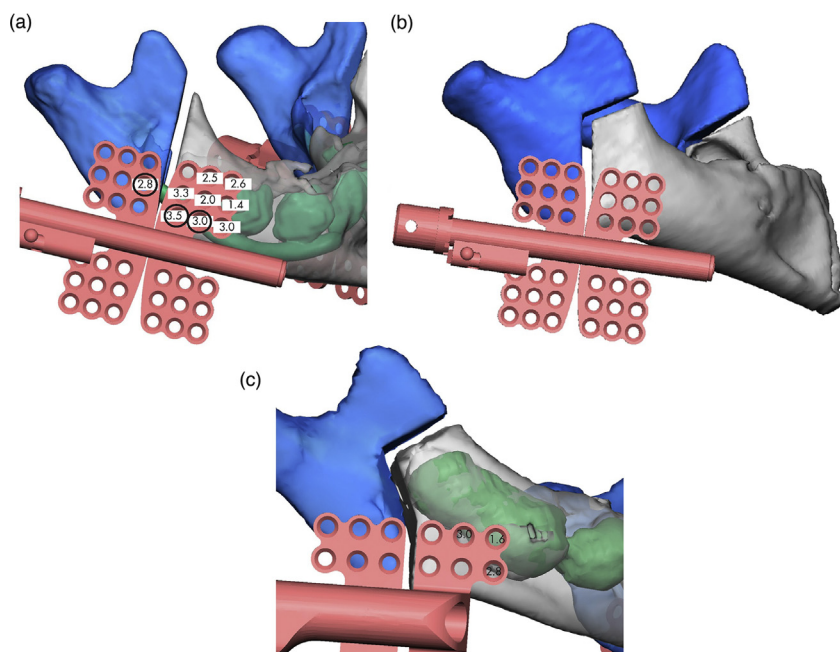


Fig. 1. Osteotomy designs for mandibular distraction. (A) A linear oblique osteotomy avoids the most posterior developing tooth (grey shadow) and overlaps the inferior alveolar nerve. Numbers indicate bone thickness from the buccal cortex to either the inferior alveolar nerve or a developing dental structure (green and grey). The screw holes outlined in black are those that are indexed in the cutting guide. (B) An inverted-L osteotomy is proximal to dental structures and ends anterior to the coronoid process. (C) A multi-angular osteotomy is necessary to avoid forming teeth (green and grey shadows) and remain anterior to the coronoid process in this patient.

Final Position Distracted 15 mm

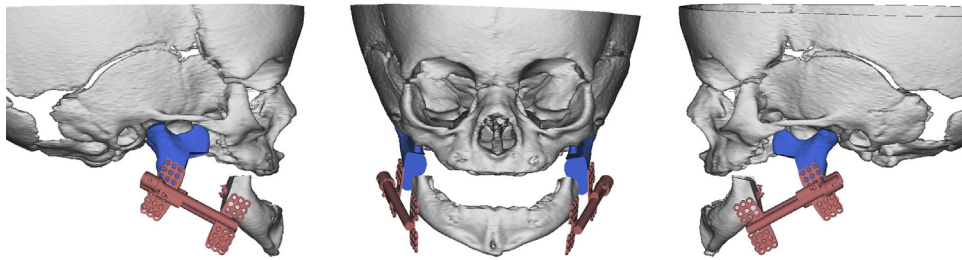


Fig. 2. Simulated distraction to 15 mm.

to be used was applied to each side of the mandible and distraction of the distal segment was simulated (Fig. 2).

This author's preference is for a horizontal distraction vector that is nearly parallel to the palatal plane^{23,24}. To achieve this, the distraction devices were placed with the activation arms extending posteriorly (Fig. 3), as anterior positioning requires an oblique or vertical vector to allow the arm to lie below the inferior border of the mandible. As a result of the horizontal device orientation, there was typically insufficient available bone at the mandibular angle to allow the osteotomy to be positioned posterior to the lingula while maintaining predictable fixation of the proximal footplate. Therefore, osteotomies were planned to overlap the inferior alveolar nerve and the position of the nerve within the simulated osteotomy was noted on the planning report for intraoperative reference. Alternative device placement could allow the osteotomy to be

positioned posterior to the inferior alveolar nerve.

Next, cutting guides were designed. The guides were configured to capture portions of the mandibular inferior border and angle to provide stable registration, but with minimal material to facilitate ease of insertion. Holes were designed to correspond with screw positions. Guide slots were customized based on osteotomy design (Fig. 4). For a linear osteotomy, only the inferior portion was guided to orient the bone cut, which was then completed after guide removal. For an inverted-L, the slot extended to the inflexion point of the osteotomy. For multi-angular osteotomies, the guide slot demonstrated the first two angles and a straight edge was used to orient the third angle to minimize bulkiness of the guide.

A planning report was generated to summarize the Web session. The report included measurements from the buccal bony cortex to underlying structures

(Fig. 1A) and to the lingual cortex to aid in hole and screw length choice. Cutting guides and a to-scale mandibular model were then 3D printed (Fig. 5A). These guides and model were used to customize the device plates preoperatively to minimize operating time and ensure an accurate fit (Fig. 5B). These guides and modified devices were sterilized for intraoperative use.

Description of the operation and distraction protocol

After induction of general anesthesia and intubation, the head was turned to one side. A 1-cm incision was created within a natural skin tension line approximately 2 cm below the mandibular inferior border after infiltration with 1:100,000 epinephrine. Dissection was performed to the inferior mandibular border while monitoring for the marginal mandibular nerve. The mandibular angle, ascending ramus, sigmoid notch, and superior and inferior mandibular borders were exposed subperiosteally. The cutting guide was inserted and correct fit was confirmed by visual inspection and tactile sensation. A piezoelectric saw was used within the slot of the cutting guide to score the buccal cortex. One screw hole proximal to and one distal to the osteotomy were pre-drilled through the guide. The guide was then removed (Supplementary Material, Video 1).

The osteotomies were continued through the lingual cortex superiorly and inferiorly. The approximate location of the inferior alveolar nerve was determined based on the planning report, and the osteotomy was created only through the buccal cortex in that area. A tunnel was created in a superficial plane to the infra-auricular exit site for the activation arm, and a rubber catheter was passed back through this track. The activation arm was passed within the catheter and connected to the pre-bent distraction device. The pre-drilled screw holes were used to



Fig. 3. The activation arm extends posteriorly from the distraction device and exits inferior and posterior to the ear lobule.

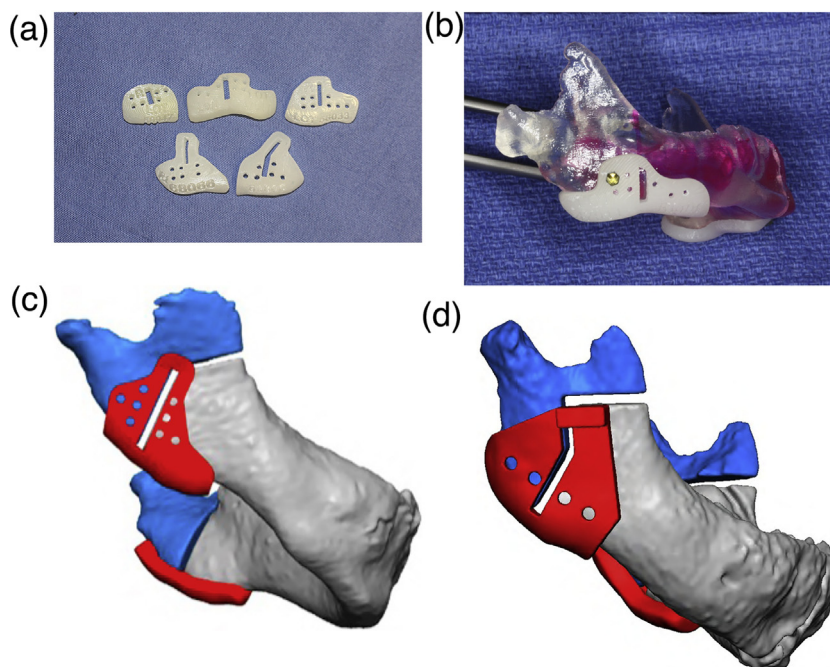


Fig. 4. (A) Examples of cutting guide designs. (B) Guides for linear osteotomies need only orient the inferior portion of the cut; the remainder can be completed after guide removal. (C) For inverted-L osteotomies, the guide can extend to the inflexion point of the osteotomy to ensure that dental structures are avoided as planned. The more horizontal limb of the osteotomy can be performed free-hand, as the angulation of this cut is less important. (D) For a multi-angular osteotomy, the guide slot dictates the two inferior angles and a flat edge can be incorporated to indicate the position and angulation of the superior limb.

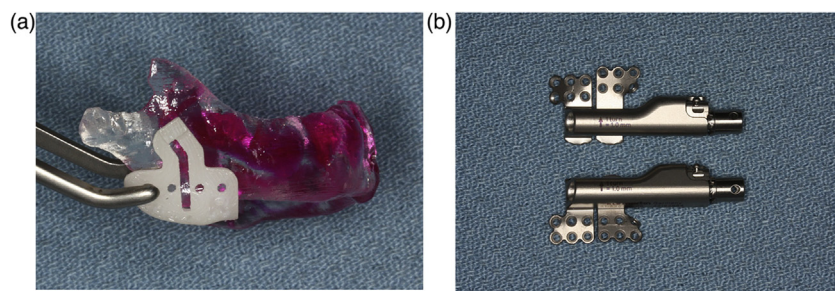


Fig. 5. (A) 3D printed cutting guide and mandibular model. (B) Distraction devices that have been modified preoperatively based on the 3D model and virtual plan.

Neonatal MDO Protocol

- Internal device with activation arm posterior
- Horizontal vector
- 2 mm intraoperative distraction
- No latency
- 2 mm distraction per day, split into 2 sessions
- Overcorrect 2-4 mm
- Intubation for 3-4 post-operative days
- Oral feeding as early as possible
- Polysomnogram at end of MDO
- Remove turning arms at bedside when MDO completed
- Consolidation for 6-8 weeks

Fig. 6. Boston Children's Hospital neonatal distraction protocol for infants with Robin sequence.

align the device, which was then secured with four monocortical screws in each foot plate.

The device was activated and the osteotomy was observed for separation. If tension was encountered, an osteotome was used to complete the osteotomy through the lingual cortex. The device was further activated to ensure tension-free separation of the segments. The device was then deactivated until a total distracted distance of 2 mm remained. The wound was irrigated and closed in layers. The operation was then repeated on the contralateral side.

After the operation, the patient was brought intubated to the neonatal intensive care unit. Distraction commenced at a rate of 2 mm per day, separated into morning and evening activations, beginning on the first postoperative day. Intubation was maintained for 3-4 postoperative days to allow airway swelling to resolve. Distraction was continued until the mandibular alveolar ridge was 2-4 mm anterior to the maxillary alveolar ridge, and then a PSG was obtained to confirm resolution of OSA. If the OSA had improved but not resolved, the devices were advanced an additional several millimeters. After completion of distraction, the activation arms were removed at the bedside. Patient disposition was then dictated by other needs, such as feeding assistance. Devices were removed approximately 8 weeks after insertion (Fig. 6).

Data analysis

Descriptive statistics were computed. A χ^2 test was used to evaluate associations between categorical variables. A P -value of <0.05 was considered statistically significant. Statistical analyses were performed using SAS version 9.4 (SAS Institute, Inc., Cary, NC, USA).

Results

Sample characteristics

The study sample included 17 patients (47.1% female). Mean gestational age at birth was 37.9 ± 2.1 weeks, and mean birth weight was 2.8 ± 0.6 kg (range 1.5-3.5 kg). Seven subjects (41.2%) had a syndromic diagnosis, including Nager syndrome ($n = 3$), Stickler syndrome ($n = 2$), skeletal dysplasia ($n = 1$), and an undefined constellation of multiple anomalies ($n = 1$). One subject (5.9%) had a tracheostomy at another institution prior to presentation to Boston Children's Hospital that remained in place during MDO,

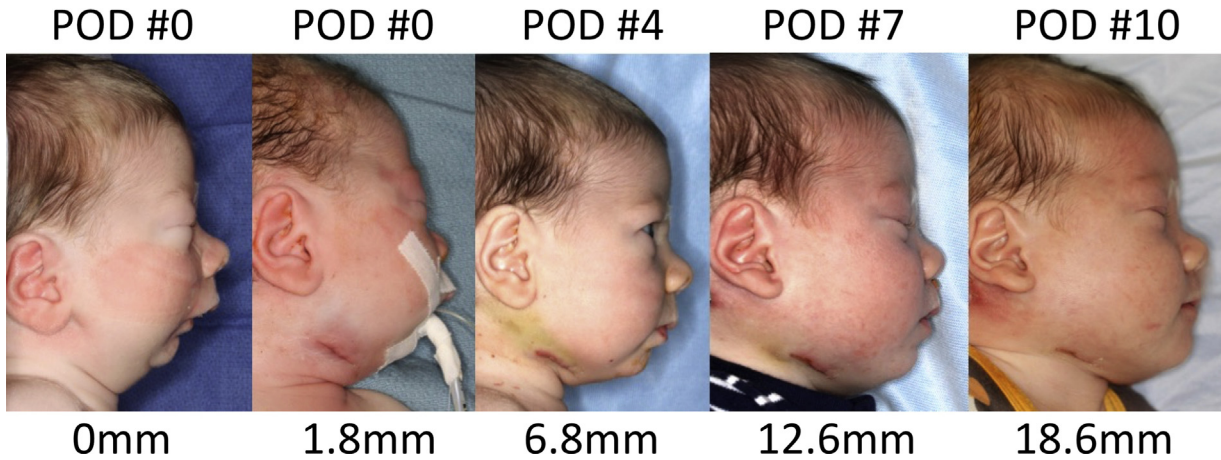


Fig. 7. Series of photographs from pre-MDO to the end of active distraction in a boy with Robin sequence who initiated MDO at age 18 days. 'POD' indicates the postoperative day at the time of the photograph; the distance distracted is indicated below each image.

and two patients (11.8%) had tongue-lip adhesions prior to MDO.

The mean age at MDO was 87 ± 96 days (range 18–302 days). The average mandibular advancement was 17.9 ± 1.9 mm (range 13.8–20.0 mm) (Fig. 7). The distraction devices were removed an average of 67 ± 15.6 days (range 39–90 days) after placement. The mean length of postoperative follow-up was 458 ± 267 days (range 26–1011 days).

Virtual surgical planning and osteotomy design

The maxillofacial CTs used for preoperative virtual planning were obtained at a mean age of 72 ± 94 days and an average of 15 ± 7 days (range 7–33 days) prior to the operation. Ten patients (58.8%) were intubated at the time of the CT, with an endotracheal tube ($n = 7$), laryngeal mask airway ($n = 2$), or tracheostomy ($n = 1$). Of these, two were already intubated because of airway distress and the remaining eight (47.1%) were intubated solely for the CT study. For one patient, the CT had to be repeated with anesthesia and intubation after a non-intubated scan was non-diagnostic due to motion artifact.

The osteotomy was designed as a linear oblique cut in four cases (23.5%, Fig. 8), an inverted-L in 11 (64.7%, Fig. 9), and with multiple angles to avoid developing dental structures in two (11.8%, Fig. 10).

Intraoperative findings

Cutting guides fit and were used as planned in all cases. After removal of the cutting guides, the marked osteotomies

were noted to mimic the virtually planned osteotomies in shape and position in all cases (Figs. 8–10).

In all cases, the distraction devices fit as registered by the pre-drilled holes made through the cutting guides. It was noted that planning for the most posterior-inferior hole of the proximal foot plate to extend off the bone behind the mandibular

angle (Fig. 1) and preoperatively bending the plate to wrap around the mandible at that hole using the 3D model (Fig. 5) facilitated intraoperative device placement and added a tactile cue for confirmation of correct device positioning.

The inferior alveolar nerve was typically preserved and visualized within the distraction gap (Fig. 11).

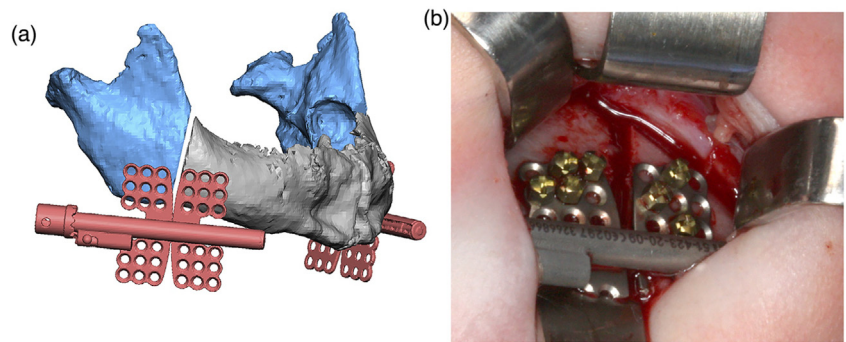


Fig. 8. Virtual (A) and actual (B) linear osteotomy.

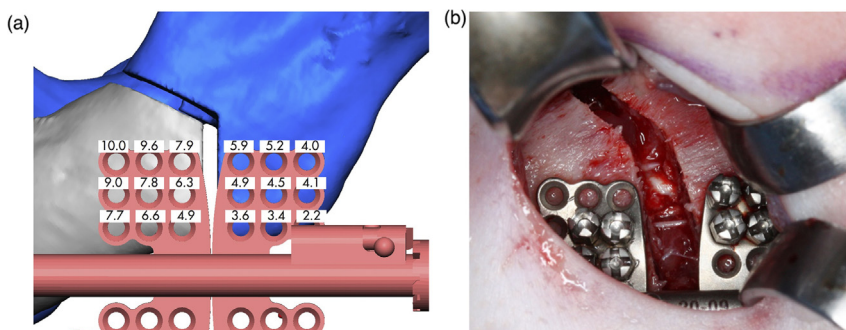


Fig. 9. Virtual (A) and actual (B) inverted-L osteotomy.

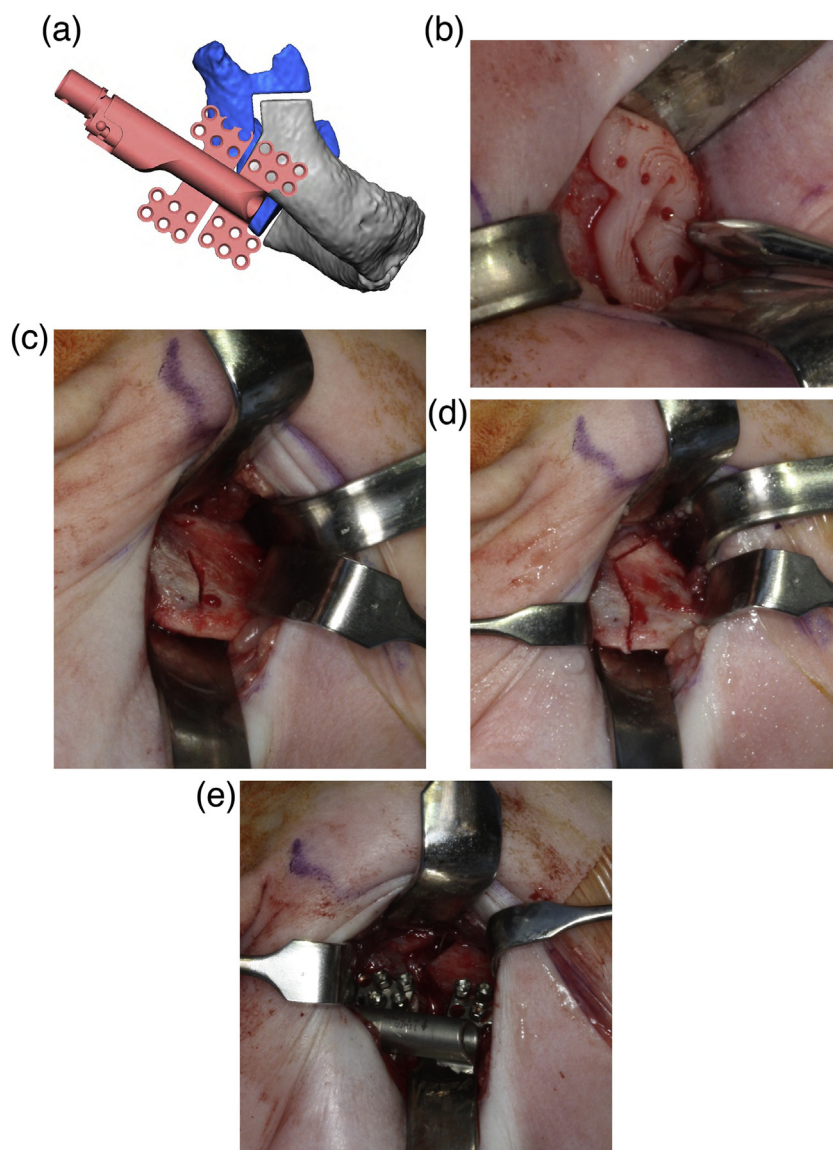


Fig. 10. (A) Simulated multi-angular osteotomy. (B) The cutting guide is placed and used to mark the osteotomy angles (two inferior and one superior) and two screw holes (one proximal and one distal to the osteotomy). (C) Marked osteotomies and indexing screw holes after removal of the cutting guide. (D) The osteotomies are completed. (E) The distraction device is applied and activated.

Outcomes

At the time of device removal, sufficient bone was found within the distraction gap to support the mandibular position without the need for additional hardware in all cases. There were no cases of screw pull-out or premature detachment of a foot plate from the bone. In some cases, bone that had grown over the edges of the foot plate or screw heads had to be removed to facilitate device removal.

PSGs were obtained for 13 subjects (76.5%) pre-MDO at an average age of 59 ± 80 days (range 3–254 days) and a mean of 22 ± 15 days (range 6–54 days)

prior to the operation. Of these, 11 subjects (84.6%) had severe OSA, one (7.7%) had moderate OSA, and one (7.7%) had mild-moderate OSA.

Postoperative PSGs were available for 15 patients (88.2%) at a mean age of 99 ± 111 days (range 27–413 days) and an average of 22 ± 34 days (range 6–141 days) after initiation of distraction. These found no OSA in 12 patients (80%) and minor residual OSA in three (20%). Two of the three patients with minor residual OSA were syndromic, but the association between syndromic diagnosis and PSG outcome was not statistically significant ($P = 0.291$). Among patients who had both

pre- and postoperative PSGs ($n = 11$), there was a mean improvement in overall severity score by 4.4 of 5 levels ($P < 0.001$).

No patients required additional airway management, including supplemental oxygen or positioning maneuvers, after MDO. Patients were discharged to home at a mean of 21 ± 16 days (range 3–72 days) after the initial operation for MDO. No patients were discharged to other care facilities. The one patient who had a tracheostomy prior to MDO was successfully decannulated postoperatively. One patient required a second MDO procedure during the follow-up period (2.6 years after the first MDO) due to recurrent micrognathia and airway distress, possibly associated with his diagnosis of Nager syndrome, and this second procedure was again successful in relieving the OSA.

There were four (23.5%) early postoperative complications that were all considered minor because they did not require a return to the operating room and did not compromise the airway result. Three (17.6%) were skin infections in the area where an activation mechanism emerged. Of these, two were managed with antibiotics and wound care alone; one required an incision and drainage procedure in the outpatient clinic. In one patient (5.9%), the foot plates were noted at the time of device removal to have separated only two-thirds of the length of the distracted distance, which was later found to be due to device malfunction. This patient nonetheless had a successful airway outcome.

There were no statistically significant associations between syndromic presentation and need for intubation for the CT ($P = 0.906$), type of osteotomy chosen ($P = 0.183$), change in PSG severity score ($P = 0.378$), or occurrence of complications ($P = 0.682$). There was no correlation between osteotomy design and complications ($P = 0.145$ for infection, $P = 0.178$ for device malfunction).

Discussion

Virtual surgical planning was first applied to mandibular distraction around the turn of the century^{25,26}. Over the subsequent decade, VSP technologies and protocols improved dramatically^{27–29}. In 2014, Doscher et al. published the first single case report applying VSP and 3D printing of cutting guides to a neonate with RS²⁰. There appears to be only one other case report of VSP techniques applied to infant MDO³⁰.

In this series of infants who underwent MDO with preoperative VSP, all subjects

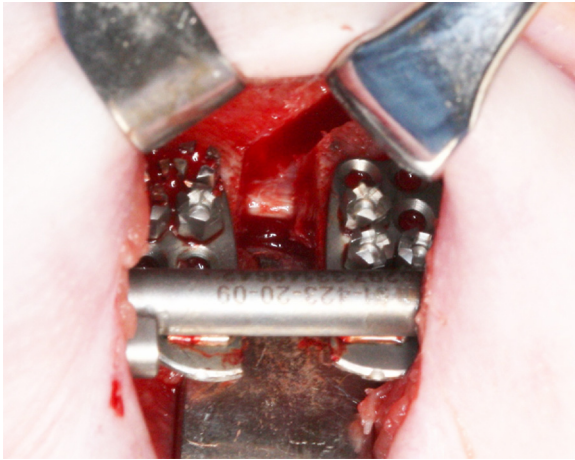


Fig. 11. Example of inferior alveolar nerve preserved within the osteotomy after the segments are distracted several millimeters.

had successful airway outcomes with complete resolution of OSA in 80% and minor residual OSA below the threshold for requiring additional airway intervention in the remainder. The one patient who had a tracheostomy pre-MDO was successfully decannulated postoperatively. Sufficient bone had formed in the distraction gaps by the time of device removal at a mean of 9.5 weeks after placement (approximately 8 weeks after completing active distraction) in all cases. No postoperative imaging was obtained to assess generated bone; timing for device removal was empiric. There were no major and four minor early complications (three wound infections managed with antibiotics and wound care; one device malfunction that still led to a successful airway outcome), and one syndromic RS patient required a second MDO during the follow-up period due to recurrent OSA.

The 3D printed cutting guides fit well and were utilized successfully to guide the osteotomy and device positioning in all cases in this series. As a result, all actual osteotomies mimicked the virtual ones. Osteotomies were customized to avoid damage to visualized dental structures and to achieve the desired vector for distraction. The location of the inferior alveolar nerve was also ascertained from the 3D modeling, which, in combination with meticulous operative technique, facilitated nerve preservation. The use of cutting guides and the preoperative bending of distractor foot plates decreased the need for wide tissue dissection, allowed fast and accurate recognition of the correct device position to achieve the planned distraction vector, aided in the identification of adequate supporting bone for screw placement, and shortened the operative time.

One potential benefit of VSP is the avoidance of damage to developing dental structures. In a series of 85 patients who underwent MDO in infancy and had a median follow-up of 7.3 years, Steinberg et al. found that 47.7% had abnormal formation of the permanent mandibular first molar tooth, and the primary mandibular second molar was injured in 13.6%.¹ The ability to modify these outcomes by 3D visualization of the developing dentition and customization of osteotomy designs, however, is unclear. The examination of skeletal remains of infants has shown that developing structures of the permanent mandibular first molar are never seen at birth and rarely found at age 1.5 months. By 4.5 months, initial cusp formation of these teeth is typically observed, and by 7.5 months, development of the crown has been completed. Root formation is not observed until 3.5 years. For the deciduous dentition, crown development of the primary mandibular first molar is usually complete and initial cusp formation of the primary mandibular second molar is seen at birth.³¹ The CT of an infant, therefore, likely reveals developing structures of the primary but not the permanent dentition. It follows that an osteotomy designed to avoid damage to the visualized dental structures may fail to preserve the permanent teeth, whose location cannot yet be determined. Nonetheless, as malformation of the primary molar can cause eruption disturbance of the succedaneous premolar, averting damage to these teeth may be advantageous.

There are some disadvantages to the use of VSP in this population. First, exposure to ionizing radiation is necessary to obtain the CT scan that is fundamental to the VSP process. Increasing evidence supports an

association between radiation exposure and cancer risk, and infants are especially susceptible^{32,33}. While the benefits of VSP alone may outweigh these risks, appropriate screening for other anomalies that may be present in a baby with RS provides additional justification for this exposure. Postoperative imaging is not routinely obtained at the author's center, in order to minimize cumulative radiation exposure. Some authors have reported promising results with magnetic resonance imaging (MRI), which does not utilize ionizing radiation, for virtual modeling and planning^{34,35}, and this may become an alternative to CT in the future. Interestingly, MRI has even proved useful for 3D modeling of osseous structures for which this modality has typically been felt to be inferior to CT³⁶, but these techniques have not yet been used in craniomaxillofacial surgery.

Another disadvantage of the VSP process is the frequent need for anesthesia and intubation to obtain the CT imaging. In this sample, 47% of subjects who did not otherwise require intubation received anesthesia and intubation to ensure a motion-free CT, and one patient who was initially not intubated required a second CT with anesthesia due to motion artifact in the first image. The process for anesthesia and intubation, particularly in infants with tenuous airways, may pose additional risk. Furthermore, if MRI were to replace CT for this indication in the future, the need for anesthesia and intubation would likely increase due to the longer duration of the MRI scan.

The use of preoperative planning may also increase the delay between diagnosis and operation. In this sample, the mean time from the CT to operation was 15 days, and the fastest turnaround was 7 days. Improvement in and increasing availability of 3D planning and printing techniques may decrease this delay in the future.

There are several limitations to this study. First, the retrospective design and small sample size limit the ability to draw conclusions from this sample. The mean follow-up of 1.2 years precludes the evaluation of long-term outcomes including dental development; follow-up for these patients will continue. Additionally, the use of PSG to evaluate MDO outcomes is complicated by the variable expression of OSA in infants and the significant airway changes that occur naturally over the first year of life^{37,38}.

In conclusion, virtual surgical planning and the fabrication of 3D printed cutting guides facilitate the precise design and

execution of mandibular distraction osteogenesis in infants with Robin sequence. Early airway outcomes are excellent. Further study with longer follow-up will be necessary to determine the influence of these techniques on long-term outcomes.

Funding

None.

Competing interests

None.

Ethical approval

This study was approved by the Institutional Review Board of the Center for Applied Clinical Investigation at Boston Children's Hospital (Protocol #P00023888).

Patient consent

Written patient consent was obtained for all identifiable photographs.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.ijom.2017.07.020>.

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