

Transfacial Two-pin External Mandibular Distraction Osteogenesis: A Technique for Neonatal Airway Obstruction from Robin Sequence

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Background: Surgical management in those with moderate-to-severe airway obstruction includes tongue-lip adhesion, tracheostomy, and/or mandibular distraction osteogenesis. This article describes a transfacial two-pin external device technique for mandibular distraction osteogenesis, utilizing minimal dissection.

Methods: The first percutaneous pin is transcutaneously placed just inferior to the sigmoid notch parallel to the interpupillary line. The pin is then advanced through the pterygoid musculature at the base of the pterygoid plates, toward the contralateral ramus, and exits the skin. A second parallel pin is placed spanning the bilateral mandibular parasymphysis distal to the region of the future canine. With the pins in place, bilateral high ramus transverse corticotomies are performed. Using univector distractor devices, the length of activation varies, with the goal of over-distraction to achieve a class III relationship of the alveolar ridges. Consolidation is limited to a 1:1 period with the activation phase, and removal is performed by cutting and pulling the pins out of the face.

Results: To guide optimal transcutaneous pin placement, transfacial pins were then placed through twenty segmented mandibles. Mean upper pin (UP) distance was 20.7 ± 1.1 mm from the tragus. The distance between the cutaneous entry of the UP and lower pin was 23.5 ± 0.9 mm, and the tragon-UP-lower pin angle was $118.7 \pm 2.9^\circ$.

Conclusions: The two-pin technique has potential advantages regarding nerve injury and mandibular growth, given an intraoral approach with limited dissection. It may safely be performed on neonates whose small size may preclude the use of internal distractor devices. (*Plast Reconstr Surg Glob Open* 2023; 11:e5085; doi: 10.1097/GOX.0000000000005085; Published online 15 June 2023.)

INTRODUCTION

Robin sequence (RS) affects roughly one in 8500–20,000 neonates and is most characterized through the triad of micrognathia, glossoptosis, and airway

obstruction.¹ An early event in this sequence is a congenitally hypoplastic and retrognathic mandible. The small mandible forces the tongue posteriorly (glossoptosis) and superiorly, which may result in the failure of fusion of the palatal shelves, leading to a cleft palate.¹ Glossoptosis leads to tongue-based airway obstruction (TBAO) of varying degrees of severity. RS has syndromic associations reported in up to 50% of cases including stickler, velocardiofacial (VCF), and Treacher Collins.²

Historically, TBAO was managed by initial prone positioning and, when not effective, tongue-lip adhesion. The goals of tongue-lip adhesion is to allow for natural mandibular growth to draw the tongue anteriorly, and thus clear the airway.^{3,4} When severe, tracheostomy may be performed to bypass the airway obstruction. More

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recently, techniques to directly address the causal factor in the sequence have been developed. Mandibular distraction osteogenesis (MDO) corrects mandibular hypoplasia and TBAO by actively lengthening of the mandible gradually, which in turn pulls the tongue forward in a composite fashion. Most craniofacial centers now widely use this technique as a first line of intervention to avoid tracheostomy.⁵⁻⁷

Surgical approaches to MDO have gone through many technical iterations in a relatively short period of time. McCarthy's initial clinical application sought to improve asymmetric micrognathia in patients with hemifacial microsomia.⁸ He utilized an external Risdon incision to allow for mandibular corticotomies or osteotomies, with external fixator placement using four monocortical pins.^{8,9} Denny expanded the use of MDO to avoid tracheostomy in patients with RS-related airway obstruction.¹⁰ Over time, the distractor devices moved toward miniaturization and internalization. The use of an internal device is favored by some centers to reduce the limitations and morbidities associated with an external device.¹¹⁻¹³ An external skin incision is typically utilized to gain access to the neonatal mandibular ramus and body. These small linear or curvilinear incisions are typically placed 1 cm inferior to the mandibular border, followed by dissection to the mandible to perform the osteotomy and place the distractor device.¹⁴ Disadvantages of this approach include a risk of facial nerve palsy or injury, excessive soft-tissue disruption, neck scars from Risdon incisions, tooth bud injury, TMJ derangement, ankylosis, and the need for a second operation to remove the device with further increased risk to the marginal mandibular nerve.^{15,16} Furthermore, the long-term effects of excessive periosteal stripping required for device placement are unknown on early bone growth.^{17,18}

One of our senior authors (C.B.G.) adapted a technique for MDO based on the transfacial pin approach described by Monasterio et al.^{19,20} The technique is highlighted by no external skin incisions, limited periosteal stripping and soft-tissue disruption, and reduced operative time. A second invasive procedure is not required, minimizing risk to the facial nerve and further facial scarring. Due to the efficacious results, this technique has been utilized over the past 17 years at our institutions. Correction of TBAO and related outcomes data using this technique have previously been described.²¹ Herein, we present our detailed surgical technique as implemented in over 150 individuals.

PATIENTS AND METHODS

Patient Workup and Surgical Decision-Making

Surgical decision-making for infants with RS has previously been published.²¹ Briefly, infants admitted to the neonatal intensive care unit with micrognathia and airway difficulties are evaluated for possible RS. This workup routinely included consults to pediatric otolaryngology, plastic surgery, speech and swallow therapy, pulmonology, and genetics. Micrognathia is assessed with physical examination including measurement of the jaw index, and

Takeaways

Question: This article reports an operative approach to the neonatal patient with severe micrognathia which has not previously been described in detail.

Findings: The technique is outlined in the context of both photographs, simulated models, and videography to allow for physician implementation.

Meaning: This article describes a technique that can be used in lower-resource settings, as the hardware requirements are not substantial and relative to standard internal mandibular distraction is particularly well suited to neonates. It has potential aesthetic advantages due to limited incisions and scar placement.

glossoptosis is evaluated at bedside using intraoral examination and nasopharyngoscopy. TBAO is diagnosed when patients have repeated desaturation (<90%) events associated with apneic episodes while positioned supine, in the setting of observed glossoptosis. When TBAO is present with micrognathia, the diagnosis of RS is assigned. Those patients then receive formal polysomnography (PSG) in a supine position, if tolerated, or side-lying or prone if not tolerated. PSG is not performed in those requiring intubation due to extreme TBAO. Surgery is considered for those with severe obstructive sleep apnea measured on PSG, generally with an obstructive index greater than 20. When surgery is indicated, a CT scan is frequently obtained to evaluate for abnormal mandibular or skull base anatomy, including asymmetric micrognathia.

Surgical Technique and Distraction Osteogenesis Protocol

Before Pin Placement

In the operating room, neonates are intubated using an oral or nasal Rae tube taped or sewn intranasally at the midline and perioperative prophylactic antibiotics are administered. Bilateral intraoral posterior mandibular buccal sulcus incisions are made to access the mandibular angles and ascending rami with minimal dissection. [See Video 1 (online), which displays the device placement. Footage demonstrates the technique of device placement.] The extent of exposure anteriorly is from the distal aspect of the most distal tooth bud to the mid coronoid process, and laterally a 1- to 2-cm region of the mid ramus, extending from the anterior to the posterior cortices.

Pin Placement

A single percutaneous 2-mm (5/64") Steinmann pin (Linear 3DX distractors, KLS Martin) is transcutaneously placed through the pre-auricular skin just inferior to the level of the sigmoid notch. The pin is advanced through the subcutaneous tissues, and under direct visualization, it is directed through the periosteum overlying the lateral border of the mandibular ramus, near the base of the coronoid process. Pin trajectory is parallel to the interpupillary line. The periosteum on the lateral and medial aspects of the ascending ramus are reflected, allowing visualization

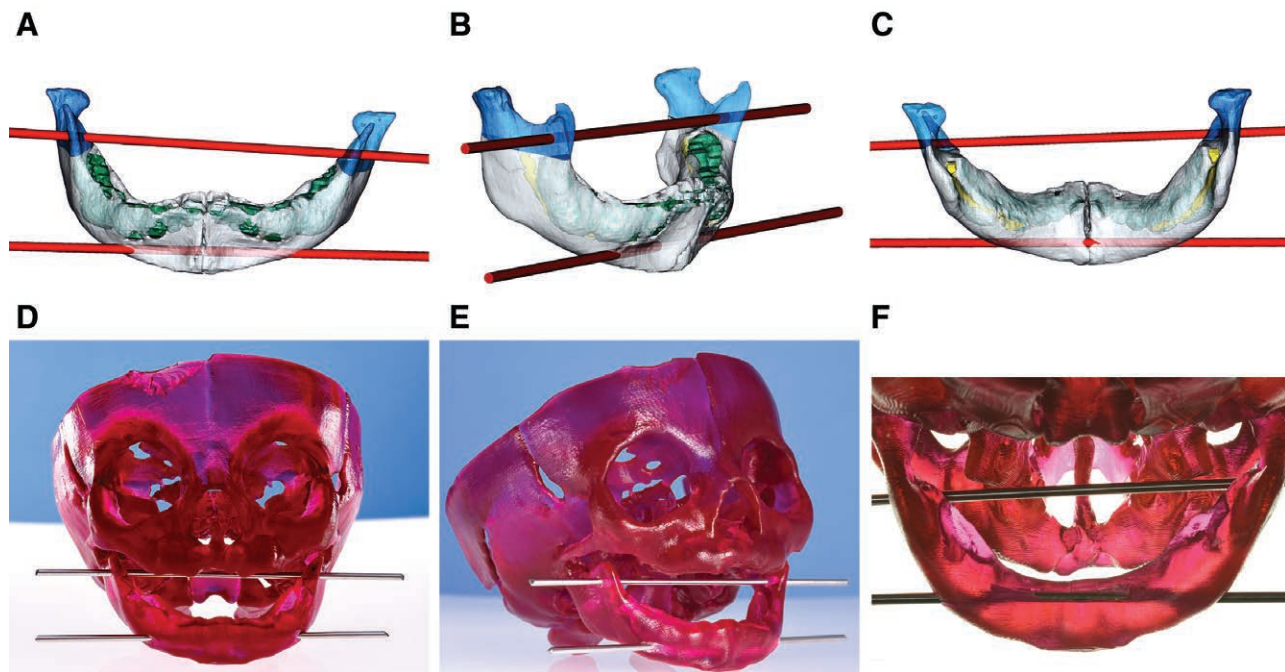


Fig. 1. Optimal pin position for the two-pin technique. Segmented CT images (A–C) and photographs (D–F) of a 3D-printed model demonstrating optimal UP and LP position. The UP is placed in the mandibular ascending ramus, within the base of the coronoid process. Care is taken to allow enough room between the pin and the most distal tooth bud for the osteotomy to be performed (along the blue/white junction in A–C). The LP is placed near the lower border of the mandibular body, inferior to all radiologically visible tooth buds, passing through all four mandibular cortices.

of the pin as it passes through the ramus. The pin is then advanced through the pterygoid musculature at the base of the pterygoid plates, toward the contralateral ramus. In patients with cleft palate, the pin may be visualized traversing the posterior oropharynx within or just cephalad to the posterior soft palate. The pin tip is again visualized as it pierces the reflected periosteum on the medial surface of the contralateral ramus. Care is taken to pierce the contralateral ramus at the same level as the initial side. The pin is visualized as it passes through the lateral cortex of the ramus, the adjacent masseter muscle, and the subcutaneous tissues and exits the skin. During the entire pin placement procedure, the surgeon and assistants communicate regarding the anteroposterior and superoinferior direction of pin placement. In case of nasal intubation, extreme care is taken to avoid injury to the endotracheal tube with the advancing pin.

A second similar pin is then placed spanning the bilateral mandibular parasymphysis/body. This pin is placed parallel to the first pin, passing through four cortices at the inferior-most aspect of the mandible to avoid damage to developing tooth buds. Pin placement is performed in a semiblind fashion. The pin is inserted transcutaneously into the body region of the mandible, just distal to the region of the future canine, and anterior to the antegonial notch. The pin is initially oriented perpendicular to the mandible as the drilling begins to allow the pin to gain bony purchase. The trajectory is then changed to run parallel to the first pin, and it is driven through both ipsilateral and contralateral mandibular cortices and out

the skin on the opposite side. The skin and subcutaneous tissues between the pins are pinched together before insertion of the second pin to allow for soft-tissue accommodation during distractor activation. Optimal pin position is demonstrated in [Figure 1](#).

Corticotomies and Distraction Protocol

With the pins in place, bilateral high ramus transverse corticotomies are performed. A piezoelectric ultrasonic osteotome (Sonopet, Stryker) is used, rather than a reciprocating saw, to prevent damage to the soft tissue and inferior alveolar nerve, given the limited incision. The corticotomies are initiated superior to the level of the mandibular foramen. As the ramus is underdeveloped, this is typically 2–4 mm above the posterior edge of the trough within the mandibular body, in which lies the most distal tooth bud.²² The osteotomy is linear, and given the patient is supine, creates a transverse or horizontal osteotomy pattern high on the mandibular ramus. The osteotomies are then completed using 4 mm osteotomes, taking care to avoid injury to the inferior alveolar nerve and the most distal tooth bud. The lower pin (LP) is then used to range the osteotomized distal mandible to ensure complete and free mobility independent of the upper pin (UP). Univector distractor devices are then placed and activated on postoperative day 1, distracting at 2–3 mm/d. The length of activation varies for each patient, but is typically at least 30 mm, and as much as 50 mm, with the goal of overdistracting to achieve a class III relationship of the alveolar ridges.^{21,23}

Extubation may be performed once the airway is cleared and confirmed with nasoendoscopy in the operating theater or ICU before total distraction is complete. Consolidation is limited to a 1:1 period with the activation phase. Neck extension positioning during the consolidation phase utilizing foam blocks assists with secretion control and prevention of airway mucosal plugging that may result from the chronically open oral aperture. A feeding tube may be necessary for nutritional support. Distractor removal is performed in the operating room (if a repeat microlaryngoscopy is needed) or bedside by simply cutting and pulling the pins out of the face. [See Video 2 (online), which displays the device removal. Footage demonstrates the technique of device removal. In this instance, removal was undertaken in the operating room; however, removal may be performed at bedside.] A “jaw bra” is placed at the time of device removal to mold the callus, closing the open bite within a few days as described previously.²³

RESULTS

Predictive Pin Placement Analysis

To guide optimal transcutaneous pin placement, we sought to localize pin exit sites with respect to surface landmarks. After obtaining IRB approval, the Wake Forest Craniofacial Imaging Database was accessed to collect facial CT scans of neonates (<3 months of age) diagnosed with RS and who subsequently underwent MDO. Patients with syndromic diagnoses including hemifacial microsomia or Treacher Collins syndrome were excluded. CT scans were then used to predict the placement of the transfacial pins, by manual segmentation of the mandibles (using Materialise Mimics software) to visualize tooth buds, the position of the inferior alveolar nerve, and the position of the overlying skin and soft tissues over the mandible. Virtual, 2-mm transfacial pins were then placed through segmented mandibles (n = 20) to maximize purchase within the mandibular bone while avoiding these structures, and the relationship of pin exit sites relative to skin and soft tissue landmarks was noted and averaged among all infants’ scans (Fig. 2). Mean UP distance was 20.7 ± 1.1 mm from the tragus for the 40 hemi-mandibles. The distance between the cutaneous entry of the upper and LPs was 23.5 ± 0.9 mm, and the tragon-UP-LP angle was 118.7 ± 2.9°. See Table 1 for summary of optimal pin placement data based on surface landmarks.

Investigation of Pin Position over Time

Serial ultra-low dose CT scans were obtained on a single patient to investigate position of the UP and LP throughout the distraction process. [See figure, Supplemental Digital

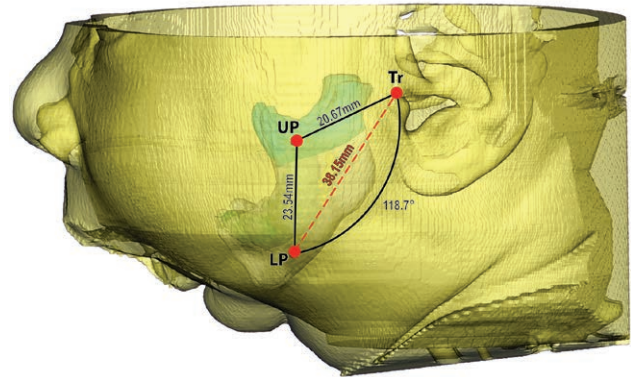


Fig. 2. Surface landmarks for pin position. Mean UP and LP positions for infants (≤3 months old), in relation to the tragon (TR). See text for details.

Content 1, which displays serial CTs in one patient undergoing two-pin technique, demonstrating pin location, mesial mandibular segment motion, and UP migration. Frontal (A–E), oblique (F–J), and inferior (K–O) views of segmented 3D-reconstructions of serial CT scans obtained preoperatively (A, F, K); at two-pin technique POD#1 (B, G, L), POD#27 (C, H, M), and POD#35 (D, I, N); and at 2 years following surgery (E, J, O). This patient had a long distraction length (45–50 mm) given their syndromic status and severe airway obstruction. With shorter distraction lengths (30 mm), the UP typically migrates cephalad through the coronoid process, resting on the pterygoid plates throughout the activation phase. Occasionally in those with greater activation lengths (and in this patient), the UP may migrate more posteriorly on the skull base, near the glenoid fossa (see G, H, L, M), <http://links.lww.com/PRSGO/C620>.]

This series of scans demonstrated upward migration of the UP to rest on the pterygoid plates, early within the activation process. Distraction of the distal osteotomy segment away from the temporomandibular joint was observed on the patient’s left side, late within the activation period. Following the device removal, both condyles were found to be appropriately positioned, and the mandibular body was significantly lengthened.

DISCUSSION

For patients with severe TBAO secondary to RS, numerous studies have corroborated the contention that MDO is among the most effective approaches available to the craniofacial surgeon to alleviate the obstruction and allow for long-term improvement in airway patency.^{10,20,24–34} A limitation to the technique remains a relatively high rate of complications. In their systematic

Table 1. Pin Placement Using Surface Landmarks

	Distance from UP to Tragon	UP to LP Distance	Tragon-UP-LP Angle
Mean	20.67 mm	23.54 mm	118.71°
SD (95% Confidence interval)	3.59 (19.55–21.78)	2.95 (22.61–24.46)	9.28 (115.76–121.66)

review, Tahiri et al identified a 23.8% complication rate associated with MDO, with severe complications including facial nerve injury and TMJ ankylosis.³¹ The authors further specified that different craniofacial centers vary regarding the preferred means of performing MDO to effectively clear the airway obstruction while avoiding complications. This review and other studies suggested that modern MDO techniques have popularized the internal device approach in an effort to decrease complication rates associated with external distraction device placement.^{10,24,27,30} Despite the overwhelmingly convincing evidence of efficacy, there are those who still criticize the use of MDO in RS, noting concerns of the complexity and morbidity of the operation, high cost, device failure, nerve and tooth injury, and growth disturbance to the mandible.^{3,35–39}

This article describes an alternative approach to MDO, eliminating external skin incisions, dissection near the marginal mandibular nerve and extensive periosteal disruption due to internal device placement. This two-pin transfacial technique further differs from the standard approach by utilizing an intraoral incision and minimal exposure of the mandibular ramus for high osteotomies. The two-pin technique is radically different than nearly every aspect of standard approaches, including the extent of soft-tissue dissection, level of osteotomy, vector of distraction, and risk to motor and sensory nerves.

Steinberg et al identified a 15% rate of lower lip depressor weakness in older children having received MDO using Risdon incisions as infants.¹⁶ The marginal mandibular nerve is at risk for injury during this procedure, at the time of both device insertion and, particularly, removal when dissecting in a scarred bed. Internal device placement requires wide soft-tissue dissection and periosteal stripping. There is evidence that aggressive periosteal disruption may lead to growth disturbances as seen with other aggressive maxillofacial dissection during palatoplasty.¹⁷ Peacock et al demonstrated decreased rates of mandibular growth in patients with RS undergoing MDO with internal devices relative to normal controls.⁴⁰ With these risks in consideration, the two-pin technique has theoretical advantages over approaches for internal devices. Small intraoral incisions allow adequate exposure of the mandibular ramus, needed only for the osteotomy. A high horizontal osteotomy has also been demonstrated to have the least risk for virtual damage to the inferior alveolar nerve compared with other neonatal mandibular osteotomy techniques.⁴¹

Another advantage of the two-pin technique is the “smaller” hardware burden and, hence, no postoperative antibiotics are needed. We have anecdotally observed higher incidences of infection when performing internal distraction, sometimes necessitating premature distractor removal. This influences some surgeons to administer long-term antibiotics postoperatively, with the associated cost and morbidity, as infection rates for internal devices in mandibular distraction osteogenesis have been found to range from 7% to 12%.^{42–44}

One concern for MDO utilizing external devices is that the associated vertical vector of distraction may load the

condyles, increasing the risk for TMJ-associated pathology, including ankylosis.^{10,23,45} Horizontal vector-based techniques have been shown to have no pathologic effects on neonatal condylar development.⁴⁶ The vector of distraction using the two-pin technique is oblique, including both a horizontal and more predominately vertical component. Based upon one patient with serial imaging and anecdotal examination findings of UP location we posit that the condyles are likely unloaded during the activation phase as the UP migrates through the thin bone of the coronoid process to rest on the pterygoid plates. This is typically clinically evident in a slight cephalic shift of the UP midway through the activation phase, and is demonstrated radiographically in **figure, Supplemental Digital Content 2**, which displays the post op, intermediate, and 6-year follow-up. Lateral images of male patient following surgery (3a and d), at intermediate follow-up (3b and e), and at 6 years (3c and f) following the two-pin technique. Results demonstrate adequate scar maturation as well as mandibular growth, <http://links.lww.com/PRSGO/C621>. However, further evaluation of postoperative CT images may be helpful to clarify UP migration over time.

An important contrast between the two-pin technique and traditional techniques is a decreased overall operative time. This approach in the hands of an experienced surgeon takes around 30–60 minutes to complete and may only require one operative trip under general anesthesia. Additionally, the total time for mandibular expansion and hardware removal is lessened, with some centers limiting long latency periods all together. This has important financial considerations for many institutions. Two-pin distractor activation is performed at a minimum rate of 2 mm/d, and we often begin at 3–4 mm/d for the first few days until patients are extubated to rapidly clear the airway. It has been the authors’ experience that a more traditional rate of 1 mm/d leads to premature consolidation when using the two-pin technique. The discrepancy in activation rates between this technique and those using internal devices may be explained by differences in hardware rigidity; unlike with internal devices, a 2-mm distractive force between two Steinmann pins does not translate to a 2-mm separation of bone at the osteotomy gap.⁴⁷ In our practice, the total activation phase typically lasts 2 weeks. The need for any consolidation phase following a two-pin technique is not established,²³ but even when incorporating a 1:1 consolidation period, distractor removal is performed near the 4-week point from the initial operation. Parent satisfaction is anecdotally improved with the two-pin external approach as the entire mandibular lengthening process and airway clearance is completed before hospital discharge.

Drawbacks to the two-pin technique exist. The psychosocial impact on parents seeing the patient with external hardware and open mouth can be difficult to adjust to. Scarring on the face may be more readily observable early than when incisions are placed under the angle of the mandible. However, it is our experience that the scars mature quite well and are minimal at long-term follow-up (**Fig. 3**; see Video 1, which demonstrates photography at



Fig. 3. Photographs of representative patient undergoing the two-pin technique. Anterior (A) and lateral (B and C) photographs of infant with RS directly following the two-pin technique. D–F, Anterior and lateral views three months postoperatively demonstrating surgical outcome.

follow-up). There is the perception that the two-pin technique has a steeper learning curve as compared to traditional MDO techniques utilizing an external approach. This is largely due to apprehension with UP and LP placement. As demonstrated in this technique article, the UP may be placed under direct visualization through an intraoral incision, and the virtual simulation should serve as a reference to identify facial anthropometric measures for LP placement in infants with RS.

CONCLUSIONS

The two-pin technique is a useful alternative to traditional internal device MDO techniques for correction of severe airway obstruction in neonates with RS. The two-pin

technique has potential theoretic advantages regarding nerve injury and mandibular growth, given an intraoral approach with limited dissection. The two-pin technique has proven to be highly efficacious for correcting severe TBAO in patients with micrognathia, and has become our standard for treating infants with RS. It may safely be performed on neonates whose small size may preclude the use of internal distractor devices.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

PATIENT CONSENT

The patient's guardians provided consent for the use of their images.

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