Treacher Collins syndrome is an autosomal dominant condition affecting the first and second branchial arches and occurring in one in 50,000 live births. The phenotype ranges from mild presentations requiring minimal medical intervention to severe anomalies requiring multiple procedures. Severe presentations of Treacher Collins syndrome have a clockwise rotation of a hypoplastic maxillomandibular complex. These manifest clinically as an anterior airway compression that is not addressed by isolated mandibular distraction osteogenesis. Our purpose is to present a surgical technique—counterclockwise craniofacial distraction osteogenesis—that improves airway morphology and occlusal rotation in tracheostomy-dependent patients with this condition.

Methods: All patients underwent subcranial Le Fort II osteotomies with simultaneous mandibular osteotomies, followed by coordinated maxillomandibular distraction with counterclockwise rotation. We reviewed pretreatment, posttreatment, and end-treatment cephalograms. Airway changes were assessed using polysomnography, sleep endoscopy, and direct laryngoscopy. Bivariate statistics were computed to compare pretreatment and posttreatment measures.

Results: Five subjects (age range, 4.5 to 12.1 years) underwent this new procedure; three had previously undergone mandibular distraction. The average palatal plane rotation was 17 degrees, the effective mandible length increase was 18 mm, and the facial plane relative to skull base rotation was 14 degrees. There was a symmetric 30 percent relapse of rotation with maintained occlusion in the first 9 months of follow-up that then stabilized. Four patients were successfully decannulated following counterclockwise craniofacial distraction osteogenesis following polysomnography. Sleep endoscopy available on two patients demonstrated resolution of the upper airway obstruction.

Conclusions: Counterclockwise craniofacial distraction osteogenesis provided greater palatal rotation than previous techniques. The resulting improvement in airway anatomy allowed for decannulation in four of five tracheotomized patients. Stability of the counterclockwise rotation is comparable to that of related orthognathic operations, despite substantially greater magnitude. (Plast. Reconstr. Surg. 142: 447, 2018.)

Clinical Question/Level of Evidence: Therapeutic, IV.
open bite; steep facial, occlusal, and mandibular plane angles; long anterior face height; and airway obstruction from both retrognathia and compression. The prevalence of obstructive sleep apnea associated with Treacher Collins syndrome dysmorphology has been reported to be as high as 95 percent, with a tracheostomy rate of 6 to 41 percent.

The goals of surgical reconstruction of the craniofacial skeleton in Treacher Collins syndrome are multifold: expansion of the upper airway, restoration of functional occlusion, and improvement in facial aesthetics. Achieving these goals in complex Treacher Collins syndrome cases remains elusive, with a high skeletal relapse rate after orthognathic surgery and mandibular distraction osteogenesis, and an inability to treat obstructive sleep apnea or remove the need for a tracheostomy despite multiple surgical attempts in many patients. In addition, there is no evidence in the literature for a reduction in the severity and prevalence of airway obstruction in Treacher Collins syndrome with age, emphasizing the need for successful early treatment.

Airway volume analysis of Treacher Collins syndrome patients has demonstrated that both maxillomandibular hypoplasia and clockwise rotation contribute to the degree of obstruction. Although the importance of the clockwise rotation deformity is well recognized, the current standard treatment of airway compromise in the immature skeleton is isolated mandibular surgery using distraction lengthening or genioglossus advancement by means of genioplasty. Both techniques are significantly limited in their ability to address the maxillomandibular rotation deformity, as they offer little to no significant counterclockwise rotation.

Counterclockwise rotation will not only correct the occlusal deformity, but is known to increase posterior airway dimensions and should be a primary treatment goal. Tulasne and Tessier recognized the importance of this and, in 1986, proposed the “procedure integral,” which attempted a correction of the rotation deformity with subcranial osteotomies, impaction, and bone grafting in one or two stages. Although a novel approach, there were multiple shortcomings that prevented widespread acceptance of the technique: a high rate of relapse of mandibular position in the immature skeleton (as early as 1 year after surgery), instability of the maxillary occlusal rotation, the surgical challenge of overcoming high soft-tissue resistance to achieve en bloc movement, and the need for extensive bone grafting.

In this report, we present a novel surgical technique that revisits subcranial rotation in Treacher Collins syndrome patients through simultaneous Le Fort II and mandibular distraction around a nasofrontal pivot. We hypothesized that counterclockwise craniofacial distraction osteogenesis (C3DO) would create a normalizing rotation of the palatal plane and result in favorable changes along the entire upper airway. With regard to this hypothesis, our specific aims were to (1) assess the cephalometric changes that occur as a result of the counterclockwise craniofacial distraction osteogenesis technique and (2) evaluate sleep study and airway data following counterclockwise craniofacial distraction osteogenesis to assess airway improvement.

**PATIENTS AND METHODS**

**Study Design/Sample**

This was a prospective study of children with a complex phenotype of Treacher Collins syndrome who underwent counterclockwise craniofacial distraction osteogenesis performed by the primary author (R.A.H.) for treatment of severe airway obstruction. All patients already had tracheostomies in place at the time of surgery. Three patients had gastrotomy tubes before surgery. The other two had previous gastrotomy tubes that were replaced at the time of surgery to help with nutrition after surgery. All patients had preoperative, end-distraction, end-consolidation, and end-treatment (at a minimum of 6 months after consolidation) lateral cephalograms. The study was approved by the Institutional Review Board of Seattle Children’s Hospital and conformed to the Declaration of Helsinki.

**Surgical Technique**

We performed a Le Fort II subcranial separation through a coronal incision. After removing 5 mm of bone below the nasofrontal osteotomy, we created a fixed point of rotation for the subcranial en bloc rotation movement during distraction.
with a 26-gauge wire hinge across the nasofrontal osteotomy. Through preexisting Risdon incision scars, we used custom virtual surgical planning cutting guides (VSP; 3D Systems, Littleton, Colo.) to mark inverted-L ramus osteotomies and guide two pairs of parallel transfacial 0.078-inch steel pins on each side of the osteotomy. This allowed accurate placement in these hypoplastic mandibles relative to surrounding teeth and nerve. Paired multivector external mandible distraction devices (KLS Martin, Jacksonville, Fla.) were attached to the pins before completing the osteotomy. We placed the patient in maxillomandibular fixation around a custom acrylic occlusal splint with extraoral traction posts from an embedded facebow. The maxillomandibular fixation was maintained with transpalatal and circummandibular 26-gauge wires secured to the acrylic splint. After closing the incisions, we applied an external midface distraction device (DePuy Synthes CMF, Paoli, Pa.) and wired the activation arms to the splint traction posts with a 45-degree upward vector relative to the Frankfort horizontal.

At the end of the operation, the subcranial facial skeleton had been mobilized and wired together as one unit with a rotation point at the nasion. We performed lateral canthopexies to the lateral orbital rims. The midface distraction device traction on the maxillomandibular fixation splint exerted a counterclockwise rotational pull on the face (Fig. 1). The mandible distraction devices were used to maintain the contact of the condyles with the skull base during the rotation, but care was taken to ensure the maximum tension was on the midface device wires and not on the mandibular pins to avoid undue compression at the temporomandibular joint.

Patients remained in the intensive care unit for 48 hours with heavy sedation. Activation of the mandible and midface distraction devices was started on postoperative day 5 at 1.5 mm/day. Patients were followed with lateral cephalograms until the palatal plane approached a normal value relative to the sella-nasion plane of 7 degrees. In patients 4 and 5, rotation was stopped short of this goal because of developing enophthalmos.

After completion of activation, the patients returned to the operating room for removal of the maxillomandibular fixation wires to allow independent mandibular movement during consolidation. In cases 2 through 5, at the time of device and splint removal, we performed simultaneous zygomatic reconstruction with full-thickness parietal cranial grafts using virtual surgical planning (Fig. 2). (See Video, Supplemental Digital Content 1, which demonstrates presurgical virtual

**Fig. 1.** Counterclockwise craniofacial distraction osteogenesis. **(Left)** The Treacher Collins syndrome dysmorphology includes a clockwise rotation of the occlusal plane with associated upper airway compression. **(Right)** The subcranial skeleton is separated from its connection to the skull base through a Le Fort II and bilateral mandible osteotomies. A wire hinge is placed at the nasofrontal osteotomy and the patient is placed in maxillomandibular fixation. A midface distractor is attached to the maxillomandibular fixation splint, and an external mandible distractor is placed with transfacial pins. The upward traction of the midface device creates a rotational force on the face and the mandible devices keep the mandible condyle in position. During consolidation, the maxillomandibular fixation is released to allow mouth opening.
planning, available in the “Related Videos” section of the full-text article on PRSJournal.com or, for Ovid users, available at http://links.lww.com/PRS/C864.)

Cephalometric Analysis

Low-dose protocol computed tomographic images and lateral cephalograms were analyzed using Dolphin image analysis software (Dolphin Imaging & Management Solutions, Chatsworth, Calif.) for lateral two-dimensional superimposition cephalometric analysis before surgery (preoperatively, T0), at the time of device removal (T1, end-consolidation), and 5 to 9 months after device removal (T2, end-treatment).

Standardized Direct Airway Analysis

Sleep endoscopy and direct laryngoscopy were performed on cases 3 and 5 immediately before surgery and at the time of device removal. Standardized views were captured at the oropharynx (with and without jaw thrust) using flexible sleep endoscopy under light total intravenous general anesthesia to preserve airway dynamics. Airways before and after counterclockwise craniofacial distraction osteogenesis were graded using the Chan-Parikh sleep endoscopy obstruction scale (0 to 3 scale, with 3 indicating complete obstruction) and the Cormack-Lehane laryngoscope grading scale (1 to 4 scale, with 1 being the easiest exposure and grade 4 being inability to visualize any laryngeal structures).31,32

Polysomnography

Polysomnography with capped tracheostomy was performed during the patient’s habitual sleep period in accordance with standards established by the American Academy of Sleep Medicine. Standard definitions of polysomnographic events and indices were used as described previously.31 Preoperative polysomnographs were obtained only in those patients able to tolerate capping of their tracheostomy. Post–counterclockwise craniofacial distraction osteogenesis polysomnography

![Fig. 2. Computed tomographic scan of case 2 before (left) and after (right) counterclockwise craniofacial distraction osteogenesis surgery and after consolidation zygoma reconstruction. In cases 2 through 5 (adolescent), full-thickness zygoma reconstruction across the facial clefts was performed using presurgical virtual planning. Split calvarial grafts were used to repair the craniotomy donor sites.](image)
was performed with the tracheostomy capped a minimum of 3 months after removal of the distraction device.

**Statistical Analysis**

Pooled data were iteratively entered into a statistical database program (IBM SPSS Version 24.0; IBM Corp., Armonk, N.Y.) for analysis. Descriptive statistics were computed to provide an overview of the sample. Paired samples analyses were used to compare preoperative and postoperative cephalometric data. Nonparametric methods were used, given the small sample and lack of confirmed normality within the data set. For all analyses, a value of $p \leq 0.05$ was considered significant.

**RESULTS**

Five subjects (age range, 4.5 to 12.1 years) with Treacher Collins syndrome underwent the counterclockwise craniofacial distraction osteogenesis procedure. Cases 1 and 2 had been treated previously at our center. Cases 3, 4, and 5 had received their previous care outside our institution. Active distraction ranged from 23 to 34 days. All cases had consolidation of the distracted mandible osteotomies at the time of device removal (Fig. 3). Cases 2 through 5 (adolescents with postremoval malar reconstruction) had consolidation of the midface osteotomies, whereas case 1 (preadolescent with no malar grafts) demonstrated rotational relapse of the maxilla and the need for surgical repositioning and plate fixation. No patients have required or are scheduled for revision mandible or maxillary surgery at last follow-up. Cases 2 through 5 were all successfully decannulated based on airway examination and polysomnographic results. Case 1 has not yet been decannulated, pending planned tongue base reduction surgery. Interincisal opening was comparable to the preoperative state. Complications associated with each operation are listed in Table 1.

![Fig. 3. Oblique view photographs of cases 2, 3, and 5 (left to right) before (above) and after (below) counterclockwise craniofacial distraction osteogenesis and autogenous zygoma reconstruction at device removal. No ancillary techniques such as fat grafting or lower eyelid reconstruction were performed. The periorbital changes were solely secondary to the counterclockwise craniofacial distraction osteogenesis and zygoma grafting procedures. Consent for use of photographs was not available for cases 1 and 4.](image-url)
Cephalometric Analysis

Cephalometric analysis results before and after counterclockwise craniofacial distraction osteogenesis surgery are listed in Table 2. Following counterclockwise craniofacial distraction osteogenesis, there were statistically significant changes noted (p ≤ 0.04). The mean facial plane rotation was 14 degrees forward, with an average relapse of 5 degrees at end-treatment. The mean counterclockwise palatal plane rotation was 17 degrees, with an average relapse of 5 degrees. Mandibular length increased an average of 18 mm, with a 7-mm relapse. The maxillomandibular relation was maintained with a stable A point–nasion–B point angle (p > 0.16).

Sleep Endoscopy Changes

Cases 3 and 5 were scheduled for pre–counterclockwise craniofacial distraction osteogenesis and 3-month post–device removal sleep endoscopy and graded using the Chan-Parikh classification of pediatric airway obstruction and the Cormack-Lehane grading of direct laryngoscopy view. On the Chan-Parikh scale at the tongue base, both cases went from a grade 3 (complete obstruction) to a grade 1 (0 to 50 percent obstructed), and from a grade 3 Cormack-Lehane laryngoscope exposure (epiglottis visible only) to a normal grade 1 exposure (anterior commissure visible) (Fig. 4).

Polysomnographic Changes

Available polysomnography study results are shown in Table 3. The post–counterclockwise craniofacial distraction osteogenesis studies were performed at least 3 months after device removal and with the tracheostomy capped and uncapped. Case 1 was unable to tolerate polysomnography preoperatively. Postoperatively, case 1 (preadolescent) had improvement of airway on endoscopy, but was not considered a candidate for decannulation until further airway interventions (e.g., tongue base reduction) could be performed.

DISCUSSION

Isolated mandible distraction osteogenesis is currently the standard treatment of immature children with Treacher Collins syndrome and severe airway compromise. Although mandibular distraction osteogenesis can correct the linear deficiency of Treacher Collins syndrome micrognathia, it does not correct the underlying clockwise rotational deformity that is fundamental to the complex airway compression. As such, although it can improve the airway anatomy in some children, in many cases it is not sufficient to normalize severely compromised airways, such as those in tracheostomy-dependent children. In this report, we describe counterclockwise craniofacial distraction osteogenesis to address the subcranial rotational deformity in five Treacher Collins syndrome patients. The four adolescent patients (cases 2 through 5) all underwent successful decannulation following expansion of their airway. In three of these patients, prior mandibular distraction osteogenesis had not been successful in removing tracheostomy dependence until counterclockwise craniofacial distraction osteogenesis was performed. This is consistent with recent literature in adult obstructive sleep apnea patients that identified occlusal rotational correction as the most predictive factor in the clinical improvement of obstructive sleep apnea. These data make a compelling argument for correction of not only the

Table 1. Patient Demographics and Surgical History

<table>
<thead>
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<td>12.1</td>
<td>9.2</td>
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<td>Previous airway operations</td>
<td>Tracheostomy</td>
<td>Tracheostomy; MDO</td>
<td>Tracheostomy; MDO (3×); tongue suspension; choana dilation (10×)</td>
<td>Tracheostomy; MDO; palatoplasty; pharyngeal flap</td>
<td>Tracheostomy choana atresia repair; posterior septectomy; inferior turbinectomy</td>
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<td>Activation, days</td>
<td>26</td>
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<td>28</td>
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<td>25</td>
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<td>Zygoma reconstruction at device removal</td>
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<td>Perioperative complications</td>
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<td>None</td>
<td>Check abscess (resolved with drainage); orbital abscess (resolved with removal of implant)</td>
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F, female; M, male; MDO, mandibular distraction osteogenesis.
sagittal maxillomandibular position, but also the facial and palatal plane rotation.

McCarthy et al. introduced the technique of distraction lengthening of the human mandible, and others later described its use in advancing the retruded midface. Since that time, the recognized benefits of distraction include the ability to overcome strong soft-tissue forces through gradual movement. In counterclockwise craniofacial distraction osteogenesis, we used combined mandible and midface distraction to exert a rotational force against the strong soft-tissue resistance of the Treacher Collins syndrome face. Because of the known risk of condylar resorption following mandibular distraction osteogenesis against high resistance, we used transfacial pins to use the adjacent soft tissue to stabilize the thin distal mandible segments from rotation and temporomandibular joint compression. We also initiated midface device activation first to minimize the strain on the mandible device. We have performed counterclockwise craniofacial distraction osteogenesis using internal mandible distraction devices in two patients with craniofacial microsoma. As this procedure evolves and is applied to less severe Treacher Collins syndrome phenotypes, internal mandibular distraction osteogenesis could also be considered. There are few data on the stability of isolated mandibular distraction osteogenesis in Treacher Collins syndrome, but one small series demonstrated a 21 to 33 percent decrease in effective mandible length and a 14 to 46 percent retrusion in pogonion position 1 year after mandibular distraction osteogenesis, which is comparable to our observations with counterclockwise craniofacial distraction osteogenesis. In addition, compared with the procedure integral in which mandible relapse was greater than the midface, all counterclockwise craniofacial distraction osteogenesis patients maintained their occlusal relationship as noted by a stable A point–nasion–B point angle measure.

We were unable to find other reports in the literature of a case series with an average palatal plane rotation of 17 degrees; thus, it is difficult to place the degree of relapse we observed into context. After counterclockwise craniofacial distraction osteogenesis, we observed a 27 percent palatal plane rotation, 35 percent mandible length, and 32 percentage sella–nasion–A point angle (maxillary advancement) relapse in the first 5 to 9 months after device removal (Table 2). In case 2, for which cephalometric data past 1 year after device removal

| Table 2. Cephalometric Measures before and after Counterclockwise Craniofacial Distraction Osteogenesis Compared to Age-Matched Normal Values |
|-----------------|-----------------|-----------------|
| Subject         | Relapse (%)     | T0-T1 T1-T2 T2-T0 | NV-
| Maxillomandibular relationship |                |                |         |
| Maxillomandibular length (Co-Pog), mm |                   |                  |         |
| Facial plane to SN (SN-NPog), deg |                   |                  |         |
| Facial plane rotation |                   |                  |         |
| Maxillary rotation |                   |                  |         |
| SN-palatal plane, deg |                   |                  |         |
| Relative facial height |                   |                  |         |
| U1-palatal plane, deg |                   |                  |         |
| Lower face-throat angle |                   |                  |         |
| Lower face-throat angle |                   |                  |         |
| Upper face height (N-ANS), mm |                   |                  |         |
| Lower face height (ANS-Me), mm |                   |                  |         |
| Neck changes |                   |                  |         |
| Ulterior palatal plane, deg |                   |                  |         |
| T0, preoperative; T1, end-consolidation; T2, end-treatment (>5 mo after halo removal); NV, age-matched normative value; —, not performed; N/A, not available. *Statistically significant (p < 0.05). **Statistically significant (p < 0.005).
were available, the early changes we observed stabilized and did not continue to change on later follow-up (Fig. 5). The counterclockwise rotation movement achieved with traditional Le Fort I impaction and bilateral sagittal split mandible advancement in non–Treacher Collins syndrome patients has been generally considered to be unstable.\textsuperscript{43,44} Although there are data to suggest that relatively small rotational movements are stable in the context of a healthy normal temporomandibular joint,\textsuperscript{45,46} Treacher Collins syndrome patients have abnormal temporomandibular joint morphology. The population of Treacher Collins syndrome patients would be more comparable to orthognathic surgical patients with conditions such as articular disk displacement. In this latter group, small counterclockwise rotations of 5 to 7 degrees have been described to result in a 36 percent relapse in both occlusal plane rotation and mandible position.\textsuperscript{47,48} Although greater rotational changes are achieved with counterclockwise craniofacial distraction osteogenesis, the stability appears to be comparable to similar patient groups undergoing much smaller counterclockwise rotation with conventional orthognathic approaches.

Two possible causes of the changes observed following counterclockwise craniofacial distraction osteogenesis are the forces of the surrounding musculocutaneous environment, or a change in centric relation of the mandible from temporomandibular joint instability. For future counterclockwise craniofacial distraction osteogenesis cases we are considering the following modifications: (1) surgical release of the geniohyoid muscles; (2) temporary paralysis of the pterygomasseteric sling using botulinum toxin; (3) extending the consolidation phase to 16 weeks; and (4) ensuring the condyles are well seated and mandible position.\textsuperscript{47,48}

Fig. 4. Case 3 sleep endoscopy views before (above) and after (below) counterclockwise craniofacial distraction osteogenesis. (Left) visualization of the retroglossal area from the nasopharynx. (Center) visualization of the retroglossal area from the nasopharynx with jaw thrust. (Right) visualization of the vocal cords on laryngoscopy. The patient went from a Chan-Parikh grade 3 (complete obstruction) and Cormack-Lehane grade 3 (epiglottis visible only) to grade 1 on both scales (essentially normal airway). Case 5 also had standardized endoscopy with similar findings. Pixilation of images is secondary to magnification of the fiberoptic view through the 3-mm endoscope.
against the skull base before placement of the transfacial pins.

In case 1, we performed counterclockwise craniofacial distraction osteogenesis at 4.5 years of age and it did not result in decannulation. There was a dramatic visible improvement in the upper airway, but the retroglossal area remained too narrow. In this patient, we did not perform zygoma reconstruction at device removal because of the patient’s young age, and we observed early midface rotational relapse that required open treatment with plate fixation for stabilization. It is unclear whether this relapse contributed to the less favorable airway result, or whether the patient had a more complex airway than the other patients.

In summary, we describe a procedure that revisits the large counterclockwise rotation movement in Treacher Collins syndrome patients envisioned by Tessier and Tulasne three decades ago, but has achieved increased stability through the use of coordinated subcranial maxillomandibular distraction. We have been able to decannulate four tracheostomy-dependent adolescent Treacher Collins syndrome patients with this technique. All four of these patients had undergone prior mandible distraction(s) and/or choana operations without successful decannulation before counterclockwise craniofacial distraction osteogenesis. Besides the airway benefit of the counterclockwise craniofacial distraction osteogenesis procedure, the occlusal plane rotation and airway improvement achieved appears to be stable after the first year, and the potential favorable impact on orthognathic surgery at maturity remains to be seen. No patients have required or are scheduled for revisional maxillary or mandibular surgery at last follow-up, which is different from the high revision rate described following the procedure integral. We anticipate that all patients will still require a traditional linear double-jaw operation at the Le Fort I level at maturity for occlusion and continued improvement in their airway.

**CONCLUSIONS**

Based on our early favorable experience, counterclockwise craniofacial distraction osteogenesis (C3DO) has replaced mandibular distraction osteogenesis as the first operation in tracheostomy-dependent Treacher Collins syndrome patients with a rotational deformity. We recommend that it be performed after the age of 7 years for the maximal chance at stability and airway resolution, and that it include zygoma reconstruction at the time of device removal.

**Table 3. Polysomnographic Measures before and after Counterclockwise Craniofacial Distraction Osteogenesis**

```markdown
<table>
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<td>Preoperatively</td>
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<td>Preoperatively</td>
<td>Postoperatively</td>
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<td>90</td>
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<td>AHI</td>
<td>48</td>
<td>6.9</td>
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<td>Mean SaO2, %</td>
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<td>98</td>
<td>n/a‡</td>
<td>98</td>
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<tr>
<td>Nadir SaO2, %</td>
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<td>93</td>
<td>n/a‡</td>
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<tr>
<td>Desaturation index</td>
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<td>3</td>
<td>n/a‡</td>
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N/A, not applicable; AHI, apnea-hypopnea index; SaO2, oxygen saturation; C3DO, counterclockwise craniofacial distraction osteogenesis; PSG, polysomnography.

*Pre-C3DO PSG studies were performed after all operations such as MDO that preceded C3DO. Post-C3DO PSG studies were performed with the capped tracheostomy tube in place at least 3 months after device removal. Case 1 was unable to tolerate a PSG preoperatively. Postoperatively, case 1 had improvement of airway but was not considered a candidate for decannulation.†Case 3 was unable to tolerate capped pre-C3DO PSG.‡Case 4 did not have pre-C3DO PSG performed.
ACKNOWLEDGMENTS

The authors would like to acknowledge Joseph Losee, M.D., and Jesse Goldstein, M.D., for providing images of their patient (case 3) for this article, and for coordinating and acting as co-surgeons with the primary author (R.A.H.), at Pittsburgh Children’s Hospital.

PATIENT CONSENT

Parents or guardians provided written consent for the use of patients’ images.

REFERENCES


