Mandibular Distraction Osteogenesis for Pediatric Airway Management

*Michael Miloro, DMD, MD**

Purpose: Mandibular retrognathia may cause upper airway obstruction in the pediatric patient due to tongue collapse and physical obstruction in the hypopharyngeal region. Mandibular distraction osteogenesis (DO) may be a useful treatment option to avoid tracheostomy. This study reviews 35 patients who underwent DO as treatment for concomitant jaw discrepancy and corrective airway management.

Patients and Methods: Thirty-five consecutive patients, 20 male and 15 female, with airway obstruction were evaluated retrospectively using clinic and hospital records. The mean age was 3.5 months (range, 36 weeks' gestation to 4 years). The group consisted of patients with Pierre Robin sequence, Stickler syndrome, Opitz's syndrome, Down syndrome with obstructive sleep apnea, Goldenhar's syndrome, Treacher Collins syndrome, and mandibular retrognathia. All patients had obstruction limited to the upper airway related to severe retrognathia and posterior tongue-base displacement confirmed with direct laryngoscopy. All patients underwent mandibular DO to avoid or remove a tracheostomy and allow development of speech and normal feeding. Each patient underwent bilateral mandibular corticotomies and placement of 2 percutaneous Kirchner wires and extraoral distraction devices. Following a 0-day latency, DO was performed at 3 to 5 mm per day (mean: 4 mm per day) for a mean total of 22.5 mm (range, 15-32 mm). The mean consolidation period was 28 days (range, 20-42 days). Preoperative radiographs (lateral cephalometric radiograph and/or CT scan) were obtained in all cases preoperatively and at least 3 months postoperatively for analysis.

Results: All patients experienced resolution of obstructive upper airway symptoms during the DO process. No patient required tracheostomy, and pre-existing tracheostomy devices were decannulated before DO completion. Apnea monitors failed to trigger in any patient postdistraction, and sleep studies were normal. The mean follow-up period was 9 months (range, 4-18 months). Radiographic analysis revealed the mean increase in posterior airway space was 12 mm. The mean decrease in overjet was 12 mm. Mandibular length increased a mean of 15 mm, and the sella-nasion-B point angle increased a mean of 16 degrees. DO complications included premature consolidation requiring manual refracture, hypertrophic scarring, device replacement, apertognathia with resolution within 8 to 12 weeks following device removal, and intraoral pin exposure. There were no cases of pin site infections or development of temporomandibular ankylosis.

Conclusion: Mandibular distraction osteogenesis is a viable option for the pediatric patient with upper airway obstruction due to mandibular deficiency to avoid a tracheostomy or other surgical intervention. Mandibular DO treats the etiology of the disease process and may allow for future growth.

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Upper airway obstruction in the neonatal period presents potentially devastating problems for the patient and parents; decisions regarding the most appropriate management for these patients are complicated and depend on the recommendations of the physicians and surgeons involved in the early care of the patient. The experience of the craniofacial team greatly affects the decision-making process and the ultimate outcomes for patients and families. These factors depend on the specific location where the child is born and the expertise and prior experience of the members of the consultation team in that particular region. Traditional methods of airway management may not be appropriate for all patients; further, all parents

*Professor, Department Head, and Program Director, Department of Oral and Maxillofacial Surgery, University of Illinois at Chicago, College of Dentistry, Chicago, IL.

Address correspondence and reprint requests to Dr Miloro: Department of Oral and Maxillofacial Surgery, UIC College of Dentistry, 801 South Paulina St M/C 835, Chicago, IL 60612-7211; e-mail: mmiloro@uic.edu

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should be informed appr[illegible]vailable options for treatment regardless [illegible]r local surgeon expertise.

Perinatal airway obstruction varies in severity from mild, moderate, to severe, with the highest risk of complete obstruction and mortality possibly from cor pulmonale due to respiratory failure and resultant right-sided heart failure.[1] Most commonly, the etiology of upper airway obstruction is associated with a craniofacial malformation or other anomaly, such as Pierre Robin sequence or Stickler syndrome, Treacher Collins syndrome, hemifacial (craniofacial) microsomia or Goldenhar's syndrome, Nager syndrome, or other disease processes that include mandibular anteroposterior hypoplasia, or developmental failure of growth of the mandible in utero, as a component of their disorder.[2] The variable expression of these disorders in the individual patient is responsible for the wide range of respiratory compromise that is observed clinically in terms of disordered breathing and inability to maintain appropriate levels of arterial oxygen saturation. A "jaw index" in millimeters (mm) has been assigned to these micrognathic patients according to the amount of overjet (OJ; mm) multiplied by the maxillary arch length (mm) measured from tragion to tragion at subnasale, divided by the mandibular arch length (mm) measured from tragion to tragion at pogonion. The normal jaw index is 4.2 ± 1.8 mm; because of the small denominator in the equation, the Pierre Robin, or micrognathic, patient may have a jaw index of greater than 15.3 ± 1.0 mm^3. Although the etiology of these disorders varies as well, the unifying facial characteristic is that of mandibular retrognathia; in utero, the patient with Pierre Robin sequence[4,5] has a small lower jaw that prevents tongue descent from between the palatal shelves during embryogenesis and results in the classic U-shaped cleft palate, glossoptosis, and mandibular retrognathia. The etiology of Pierre Robin sequence is speculative, and theories include a positional malformation in utero, intrinsic mandibular hypoplasia, neurologic/neuromuscular disorders, or a connective tissue disease process. The airway obstructive symptoms of the disease are present at birth in approximately 70% of affected individuals with a wide range of severity.[6] Although normal growth and craniofacial development may improve the obstructive airway symptoms within the first 4 to 6 months of age, immediate treatment in the perinatal period is directed at maintaining an inspired oxygen saturation (S_pO_2) at 90% or greater on room air for a duration of more than 90% of the time. Because of the associated cleft palate, these patients may also present with feeding difficulties requiring nasogastric (NG) tubes, gastric tubes, and specialized nipples or obturators for feeding. Speech is also affected, and the speech disturbances may be proportional to the respiratory problems.[7]

Several methods have been proposed to manage the obstructed airway in the neonatal period of the micrognathic patient depending on the degree of severity. Mild cases may be managed with positional therapy in a prone or lateral position with neck extension to maintain acceptable oxygen saturations. If tolerated by the young patient, continuous positive airway pressure (CPAP) may be used. In the moderate severity category, nasal stents using a small-diameter endotracheal tube (eg, 3.0 or 3.5) may be used to prevent the tongue from contacting and obstructing the posterior pharyngeal wall. A pediatric laryngeal mask airway has also been used for this purpose. For moderate-to-severe and severe obstructive airway patients, a surgical option is usually necessary. Tongue lip adhesion, or anterior glossopexy, was reported by Douglas in 1946.[8] Even at the time that this technique was described in the literature, the success rates were unacceptably low, and a recent study by Denny[9] has confirmed the unpredictable success of a tongue lip adhesion procedure. In this study, there was an 18% success rate, and 10 of the 11 patients required a mean of 1.9 additional procedures following the tongue-lip adhesion to address airway or feeding problems. Even with an additional mucosal adhesion technique, the success of tongue lip adhesion to alleviate airway obstruction is less than ideal.[10] A hyomandibulopexy, similar to the procedure for obstructive sleep apnea (OSA), has also been proposed to elevate the hyoid bone and open the posterior airway space; however, this procedure is rarely performed. Ultimately, for the severely compromised airway, endotracheal intubation or tracheostomy (or both) provide an immediate solution to the problem.

Although tracheostomy is an excellent immediate option to manage severe upper airway obstruction, the procedure carries significant potential complications and postsurgical sequelae. It is best to consider tracheostomy in this clinical scenario as a short-term solution to a complex problem with long-term morbidity without clearly addressing the anatomic problems responsible for the airway obstruction. The morbidity of 25% to 50% and perioperative mortality of 0.5% to 5.0% at major centers that perform these procedures in the newborn on a regular basis are unacceptably high.[11,12] There are perioperative complications such as bleeding, pneumothorax, hypotension, cardiac arrhythmias, peritracheal insertion, and death; postoperative complications such as tube occlusion, tube displacement, the development of a tracheoesophageal fistula, vocal cord paralysis, and subcutaneous emphysema; and late postdeccanulation problems such as tracheal stenosis, tracheomalacia, unesthetic hypertrophic scarring, residual stomal defect, granuloma formation, and possibly the rare occurrence of a tracheoinnominate artery fistula. In general, growth will not allow removal of the tracheostomy until 2 to 4 years of age, which affects not only psychosocial and intellectual development but also speech and language development.[13] Perhaps most disturbing to parents is the social stigma of the presence of the tracheostomy tube and the hygiene and maintenance requirements.

Since the late 1990s, the techniques of distraction osteogenesis (DO) of the maxillofacial skeleton have been applied to the neonatal population with airway compromise. There are several advantages to this treatment approach. The DO procedure treats the etiology of the problem by lengthening the mandible, advancing the base of the tongue off the posterior pharyngeal wall, elevating the hyoid bone, enlarging the hypopharyngeal region, and eliminating the airway compromise. This is not a short-term solution but a treatment option that addresses the specific etiology of the problem. The multitude of studies and small case series that have been reported over the past 10 years have described excellent results when DO is applied to this patient population with neonatal airway compromise, mandibular hypoplasia, and airway obstruction confined to the area of the base of the tongue above the level of the vocal cords. These studies have indicated that a tracheostomy could be avoided or an existing tracheostomy may be decannulated and removed during or following the DO process. Because long-term follow-up is not yet available, the issues of future growth are yet to be determined in the neonatal DO patient population. Certainly potential complications from the DO procedure performed in such a young patient population exist depending on the specific technique employed, and these complications include facial scarring, pin loosening, device failures, failure to relieve the airway obstruction or decannulate a tracheostomy successfully, aspiration pneumonitis, pin-site infections, premature consolidation of the bony regenerate, nerve injuries (eg, inferior alveolar nerve or the marginal mandibular branch of the facial nerve), injuries to developing tooth buds, temporomandibular joint dysfunction, joint ankylosis, and potential growth restrictions, but these are reported infrequently. In the end, there is no consensus regarding management of the airway problems in the Pierre Robin patient, nor is there consensus in the literature regarding standardization in reporting of the patient population or of surgeon experience and techniques. In some studies, evaluating the neonatal outcomes of Pierre Robin newborns, there were a significant number of perinatal mortalities,[14] whereas in others, no deaths occurred, and only a small number of patients required tracheostomy.[15] These discrepancies in the literature have led to further confusion regarding diagnosis, staging, and treatment of these patients.

The purpose of this study is to present data on a group of 35 consecutively treated patients with airway compromise due to mandibular anteroposterior hypoplasia managed with bilateral mandibular distraction osteogenesis. This article presents a novel technique for the surgical procedure and also discusses indications and contraindications as well as risks and benefits and possible complications of the technique. The outcomes assessment includes an analysis of the bony and soft tissue changes following DO and the cephalometric changes of the mandible and posterior airway space based on pre- and postoperative radiographs. A treatment algorithm based on the severity of airway symptoms in the neonatal period is presented.

Patients and Methods

This is a retrospective hospital record review of 35 consecutively treated patients at the University of Nebraska Medical Center, Children's Hospital of Omaha, and Creighton University Medical Center by a single surgeon (M.M.), with the assistance of oral and maxillofacial surgery residents, between October 2002 and October 2006. Inclusion criteria for the study included a history of repetitive intermittent upper airway obstruction (UAO), patients who had a tracheostomy for UAO or in whom a tracheostomy was contemplated, frequent apneic episodes with S_PO_2 values less than 70% on room air with repeat apnea monitor triggering, labored breathing or stridor, cyanosis, syndromic or nonsyndromic mandibular hypoplasia with a significant OJ relationship of the teeth or alveolar ridges (>8 mm), direct laryngoscopic documentation of intermittent complete or near complete tongue base obstruction without other laryngeal or tracheal abnormalities (eg, significant laryngomalacia and/or tracheomalacia), and otherwise healthy candidates for a distraction osteogenesis procedure following an informed consent discussion with the parents regarding the alternative treatment options and risks and expected short- and long-term benefits of DO. All patients underwent radiographic imaging with a lateral cephalometric radiograph and/or a computed tomography (CT) scan preoperatively and at least 3 months postoperatively. Certain cephalometric landmarks were identified and traced to determine changes in the hard and soft tissues. Patients were excluded from undergoing a DO procedure if the airway compromise was mild and could be controlled with positional therapy, if the central causes of apnea could be identified, or secondary airway lesions such as tracheal webs or lack of development of the laryngeal or tracheal cartilage structures were present.

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Results

PATIENT DEMOGRAPHIC ANALYSIS

Thirty-five consecutive children (20 boys and 15 girls) with UAO were evaluated retrospectively using clinic and hospital records. The mean age was 3.5 months (range, 36 weeks of gestation to 4 years). At the time of distraction, 28 of 35 patients were less than 9 months of age, and 30 patients were less than 1 month of age; 25 patients were treated at less than 1 week of age. The patient group consisted of Pierre Robin sequence (moderate to severe),[19] Stickler syndrome,[3] Opitz syndrome,[2] Down syndrome with OSA,[2] Goldenhar's syndrome,[2] Treacher Collins syndrome,[2] and mandibular retrognathia not otherwise specified.[5] All patients underwent a multidisciplinary evaluation by several individuals including a geneticist, neonatologist, pediatric pulmonologist or pediatric otorhinolaryngologist, a pediatric anesthesiologist, and an oral and maxillofacial surgeon. All patients and parents underwent an informed consent conference with as many members of the team present as possible to present all the available options and risks and benefits of the procedures. All patients demonstrated frequent apneic episodes and repetitive upper airway obstructive signs and symptoms. Apnea monitoring demonstrated frequent apneic episodes and oxygen desaturation (70%-80%) in all patients. All patients had obstruction limited to the upper airway related to severe retrognathia and posterior tongue-base displacement confirmed with direct laryngoscopy (DL) by an otorhinolaryngologist. Thirteen children were fed by gastrostomy tube, and 18 patients were fed using a Dobhoff or NG tube. Of the 35 patients, 9 had a tracheostomy in place, and a tracheostomy was contemplated in 26 patients. Three of the Pierre Robin patients had already undergone bilateral mandibular DO by another surgeon without successful resolution of their airway symptoms, resulting in a tracheostomy in 2 of those cases. Other than the tracheostomy patients, no patient had any other form of treatment before DO in an attempt to resolve their UAO symptoms. All patients had a preoperative CT scan, with 3-dimensional reconstruction, when possible, or a lateral cephalometric radiograph to document the posterior airway space (PAS), mandibular anteroposterior position relative to the cranial base (SNB), mandibular length (condylion [Co]-pogonion [Po]), and the maxillomandibular relationship (measurement of OJ between the maxillary and mandibular central incisor buds or the anterior-most aspect of the maxillary and mandibular alveolar ridges). The postoperative radiograph was taken at least 3 months following the removal of the devices for skeletal and airway analysis.

SURGICAL PROCEDURE

The surgical technique employed in all cases of mandibular distraction osteogenesis consisted of the use of external multivector distraction devices (KLS Martin, Jacksonville, FL, or Synthes Maxillofacial, Paoli, PA; Fig 1). All patients had general anesthesia using either an existing tracheostomy or an oral intubation tube, with perioperative prophylactic antibiotics. Bilateral intraoral posterior mandibular buccal sulcus incisions were made to access the posterior body angle, and lateral and medial rami of the mandible. A single percutaneous Kirschner wire or pin (5/64 inches) was drilled through the skin and subcutaneous tissues beginning in the preauricular region to pierce the periosteum overlying the lateral border of the mandibular ramus in the upper third and posterior

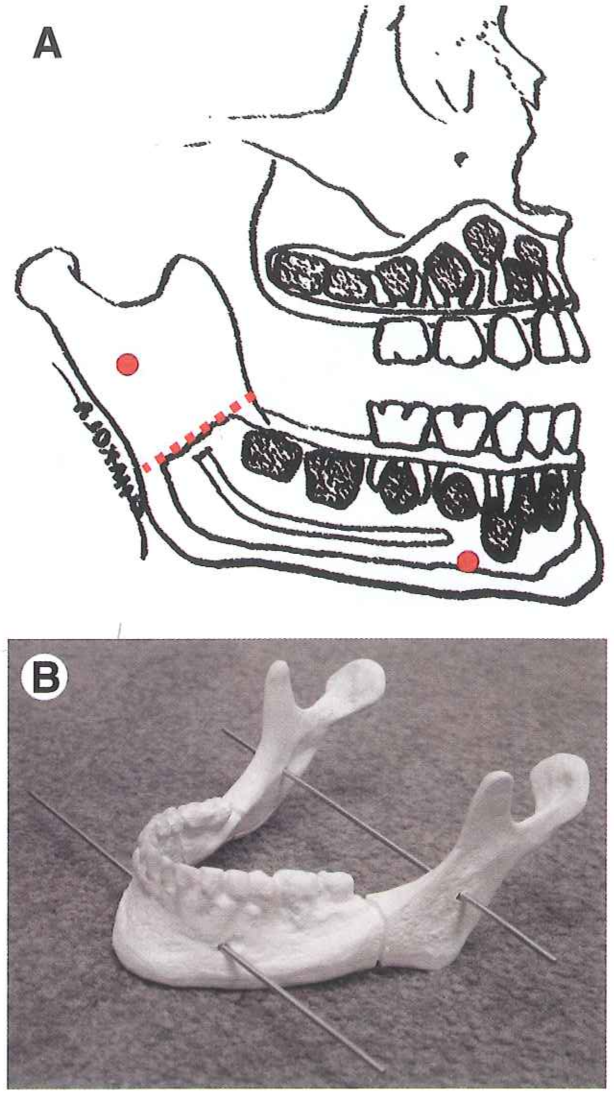

FIGURE 1. Diagram (*A*) and model (*B*) of pin placement and osteotomy locations in the mandible.

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third of the ramus (Fig 2). The direction of pin placement was parallel to the interpupillary line, when possible. The pin entrance site was in the area just superior and posterior to the midpoint of the ramus. Under direct visualization, the pin was drilled through the lateral ramus with retraction of soft tissue, and then attention was directed medially, where the medial periosteum and medial pterygoid muscle were retracted in a manner similar to that used for the horizontal cut performed during a sagittal split osteotomy of the mandible. The pin was observed to exit the medial ramus, the medial soft tissues were then

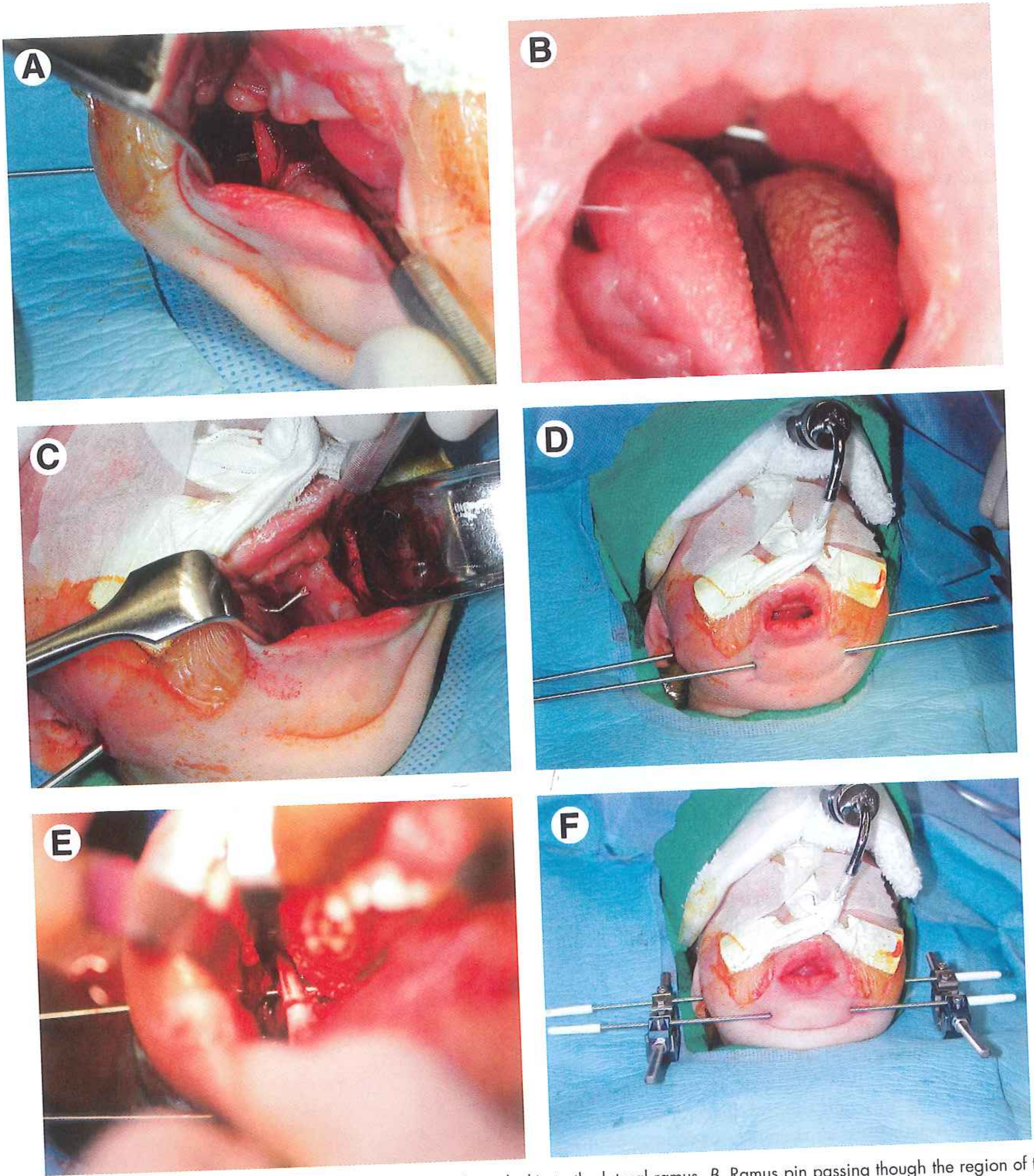

FIGURE 2. Surgical procedure. *A*, Ramus pin placement through skin to the lateral ramus. *B*, Ramus pin passing though the region of the cleft palate *C*, Ramus pin passing through the medial soft tissues through the contralateral ramus. *D*, Both pins in place. *E*, Angle corticotomy completed. *F*, Distraction device placement.

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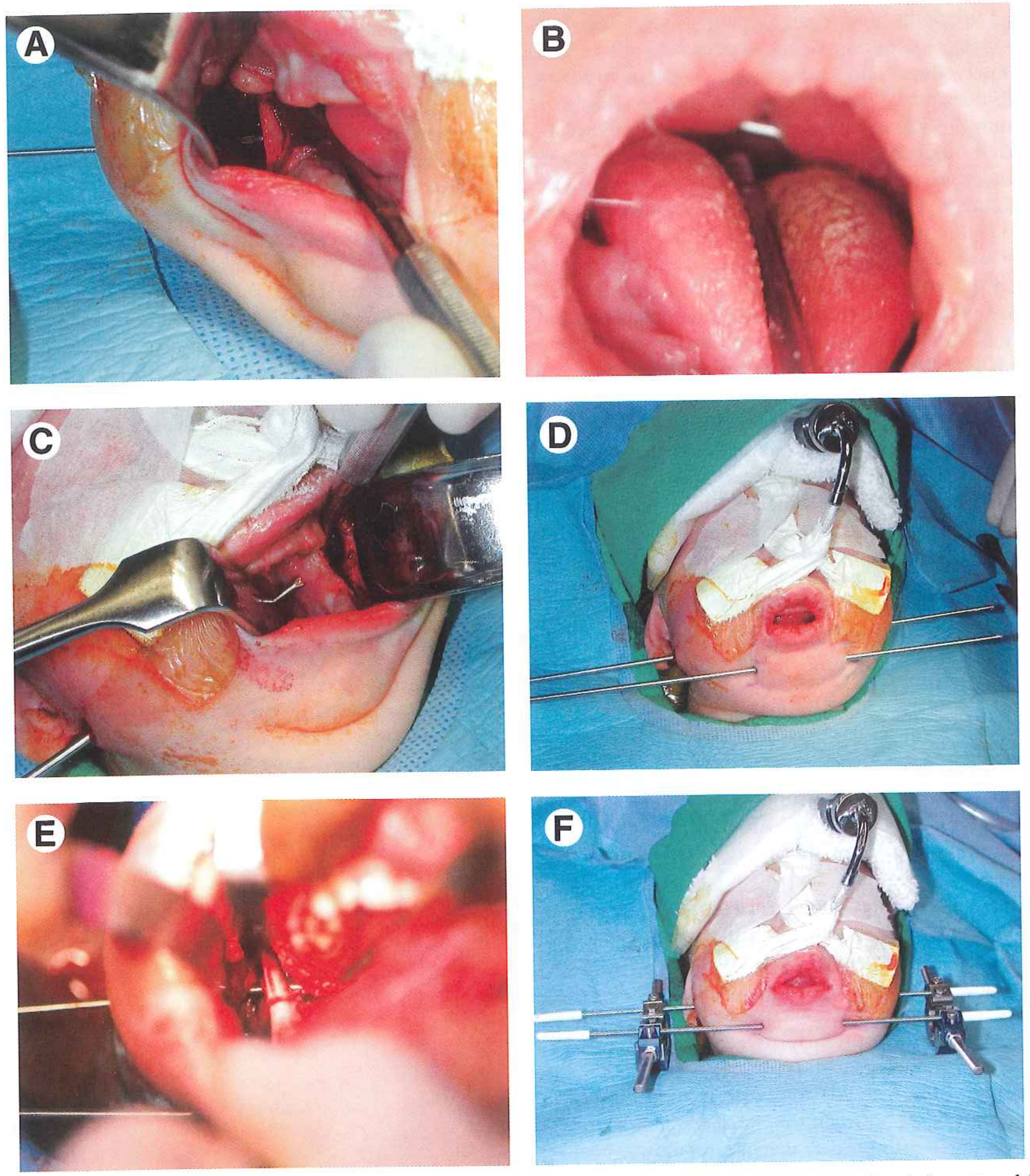

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allowed to relax, and the pin was observed to pierce the soft tissues directly. In most cases of Pierre Robin sequence, the large cleft of the palate allowed excellent visualization of the pin as it traversed the posterior oropharynx in the area of the uvula and distal soft palate. On the contralateral side, a similar approach was used, by first piercing the medial soft tissues of the medial mandibular ramus and then retracting these tissues medially to allow direct visualization of the pin as it entered the mandible in the appropriate posterior and superior location. Next, attention was again directed laterally to allow visualization of the pin exit site from the lateral ramus. Finally, the pin was drilled through the lateral periosteum and masseter muscle and subcutaneous tissues to exit the skin on the contralateral side. During the entire pin placement procedure, the surgeon and assistants communicated regarding the anteroposterior and superoinferior direction of pin placement.

In a similar fashion, a second Kirschner wire or pin (5/64 inches) was drilled through the skin in one side of the symphysis region, then through the mandible in a location that was anterior enough to avoid the inferior alveolar/mental nerve and inferior enough to avoid the developing tooth buds, especially of the most inferiorly placed canine. The area chosen for this pin placement was just superior to the inferior border of the mandible in the parasymphysis region. The direction of the pin placement was exactly the same as the first pin, with an attempt made to maintain parallelism between these 2 transcutaneous pins. Additionally, an attempt was made to "pinch" the skin and subcutaneous tissues together before insertion of the second pin to allow for soft tissue accommodation during DO. Again, under direct visualization with retraction of the mandibular buccal vestibular unattached tissues anteriorly, the skin was pierced with the pin, and under direct visualization, the pin entered the mandible in the desired location just anterior to the canine tooth bud and superior to the inferior border of the mandible. Then, in a blind fashion, while maintaining parallelism of the anterior pin with the previously placed posterior pin, the pin was advanced through the floor of mouth soft tissues to contact the medial aspect of the mandible in the contralateral parasymphysis region. Upon exit of the pin through the lateral aspect of the mandible in this region, the position was confirmed to be anterior to the canine tooth bud and close but not impinging on the inferior border of the mandible. Finally, the pin was advanced through the lateral soft tissues to exit the skin; however, before this maneuver, the skin and subcutaneous tissues were again "pinched" together under similar tension to the other side to allow for soft tissue accommodation during DO.

Next, an area in the mandibular angle region was chosen for the osteotomy, or, initially, the corticotomy, which was created in an oblique fashion. The entrance of the inferior alveolar neurovascular bundle was identified, which, in the neonatal period and in the young child is located in the area of the medial aspect of the mandible in the area that would later be occupied by the mandibular third molar tooth if it were present. As a result, the osteotomy was easily placed posterior to this landmark to avoid injury to the inferior alveolar neurovascular bundle. A reciprocating saw was used to perform the osteotomy bilaterally, taking care to ensure that the bony cuts were symmetrical and did not impinge on the location of the 2 previously placed Kirschner pins. Before completion of the osteotomy with osteotomes, the external multivector distraction devices were placed bilaterally and fixated to the pins near their exit points from the skin in 4 locations. The devices were configured to lie in a passive position with a horizontal vector consistent with the osteotomy placement in the mandibular angle region. The distraction devices used (either Synthes Maxillofacial Multi-Vector or KLS-Martin LP 3DX) were placed initially with a straight-line (or 180-degree) configuration with plans to control the superoinferior distraction vector by angular adjustment to approximately 90 degrees by the completion of the DO process. The devices were then removed, and the osteotomies were completed bilaterally. The external multivector devices were replaced and activated bilaterally to ensure movement of the bony segments to approximately 5 mm bilaterally, and then the devices were deactivated to achieve bone-to-bone contact. The intraoral wounds were irrigated and suctioned and closed with resorbable sutures. The pins were trimmed just beyond the distraction devices, and protective plastic balls were placed on the ends of the pins. The pin exit sites were treated with a triple antibiotic ointment. In some cases, mittens, Posey restraints, or both were used to prevent inadvertent or intentional grasping of the external devices. The patients were generally transferred to the neonatal intensive care units with either their previously placed tracheostomy in place or the endotracheal tube in place.

DISTRACTION PROTOCOL

Following a 0-day latency, DO was performed at 3 to 5 mm per day (mean: 4 mm per day) for a mean total of 22.5 mm (range, 15-32 mm). Whenever possible, distraction at 2.0 mm each was performed at 12-hour intervals. The patients tolerated each distraction procedure well, without signs of discomfort or distress. As distraction continued, the resistance to distraction increased subjectively because of both early bony consolidation and soft tissue restriction to the magnitude of advancement. Typically, approximately 10 days of distraction were required for 35 to 40 mm of device expansion to achieve 20 to 25 mm of forward bony mandibular advancement. The difference in these 2 numerical values results from the soft tissue restriction to the distraction vector and the inferior movement of the mandible due to resistance from the suprahyoid muscles. The endpoint for distraction was an open airway on DL, ability to breathe freely around an existing endotracheal tube, or ability to tolerate capping of the tracheostomy, if present, with a zero or negative OJ relationship of the maxilla and mandible on clinical examination during maximum intercupation of the teeth or ridges. The devices were removed following bony consolidation in the outpatient clinic without local anesthesia or sedative measures. Two of the devices were removed from the pins on one side of the patient, and the pins were cleansed with a Betadine solution. Then, the pins, connected to the devices on the contralateral side, were removed in a smooth movement without any hard tissue or significant soft tissue resistance or excessive bleeding or pain in any patient. Following a brief period of crying, the patients tolerated the pin removal procedure well without any complications.

CLINICAL AND RADIOGRAPHIC ANALYSIS

The mean follow-up period for all patients was 9 months (range, 4-18 months). The mean consolidation period was 28 days (range, 20-42 days). This amount of time was chosen empirically as an estimate of approximately 2 times the distraction period of 10 to 14 days. Clinically, all patients had improved subjective airway symptoms, before completion of the distraction period, because all patients were extubated or decannulated before the completion of the distraction process. No patient had apneic events postsurgically, and apnea monitors were discontinued within 1 week by all parents because of a lack of any alarms. Any postdistraction sleep study for OSA was normal following distraction. All feeding tubes were removed within 3 weeks after distraction, and all patients gained weight appropriate for their age. The mean intensive care unit time was 7.9 days (range, 0-20 days) based on the need for continued intubation during the distraction period. The mean hospital stay was 12.6 days (range, 9-25 days). Preoperative and postoperative (at least 3 months) radiographs were taken and compared with assessed skeletal and airway changes (Fig 3).[16] The lateral cephalogram or CT scan was marked with the usual cephalometeric landmarks by one surgeon (MM), with specific attention directed toward the PAS defined by a line from B point (B) through gonion (Go) extending through the airway shadow and a measurement made from the anterior to posterior pharyngeal wall. Mandibular anteroposterior position was defined by the angle from sella (S) to nasion (N) to B point (B) or SNB (sella-nasion-B point angle). The mandibular length was defined as the distance from condylion (Co) to pogonion (Pg). Finally, the maxillary-mandibular relationship was defined by the amount of OJ, in this case using A point and B point (because there may not be maxillary or mandibular incisors) in perpendicular relation to Franfort horizontal line (FH). Comparisons were then made between the preoperative and postoperative radiographs (Fig 4).

For a mean distraction distance of 22.5 mm, the mean increase in the PAS was 85% (4.9 mm increased to 12.0 mm). Other studies have used either multiple points in the posterior airway space or have evaluated the overall volume of the hypopharynx using 3-dimensional radiography.[17-20] The distance between the anterior and posterior airway space was measured and recorded as the PAS. The mean decrease in OJ was 12 mm (range, 10-22 mm), and most patients (32 of 35) had a reverse OJ (Fig 5) indicating a planned overcorrection to allow for either some relapse or to allow unimpeded downward and forward maxillary growth. The mean increase in SNB was 16 degrees, and the mandibular length (Co-Pg) increased by a mean of 15 mm radiographically. These skeletal changes were consistent with the planned distraction vectors and objectives of the surgical procedure and distraction protocol.

COMPLICATIONS

The most frequent complication following distraction was the development of an anterior open bite of the occlusion or anterior alveolar ridges. Of the 35 patients, 18 had apertognathia following distraction. With time, feeding, and bony remodeling, all open bites resolved within 3 months following device removal. With experience this outcome was managed preventively with the encouragement of feeding or at least nippling during the distraction phase of treatment to maintain the vertical position of the anterior mandibular segment or using an elastic head band (Fig 6). There was 1 case of failure to diagnosis laryngomalcia preoperatively, which led to a tracheostomy 12 months following completion of distraction osteogenesis. This was a curious case in which the airway was stable for a period of 1 year before redevelopment of symptoms, and direct laryngoscopic confirmation of laryngomalacia and a floppy epiglottis causing intermittent airway obstruction. There was 1 premature consolidation of the bony segments on 1 side requiring manual refracture of the bony callus. This occurred early in the primary surgeon's experience, when the rate of distraction was less than 4 mm tissue restriction to the magnitude of advancement. Typically, approximately 10 days of distraction were required for 35 to 40 mm of device expansion to achieve 20 to 25 mm of forward bony mandibular advancement. The difference in these 2 numerical values results from the soft tissue restriction to the distraction vector and the inferior movement of the mandible due to resistance from the suprahyoid muscles. The endpoint for distraction was an open airway on DL, ability to breathe freely around an existing endotracheal tube, or ability to tolerate capping of the tracheostomy, if present, with a zero or negative OJ relationship of the maxilla and mandible on clinical examination during maximum intercupation of the teeth or ridges. The devices were removed following bony consolidation in the outpatient clinic without local anesthesia or sedative measures. Two of the devices were removed from the pins on one side of the patient, and the pins were cleansed with a Betadine solution. Then, the pins, connected to the devices on the contralateral side, were removed in a smooth movement without any hard tissue or significant soft tissue resistance or excessive bleeding or pain in any patient. Following a brief period of crying, the patients tolerated the pin removal procedure well without any complications.

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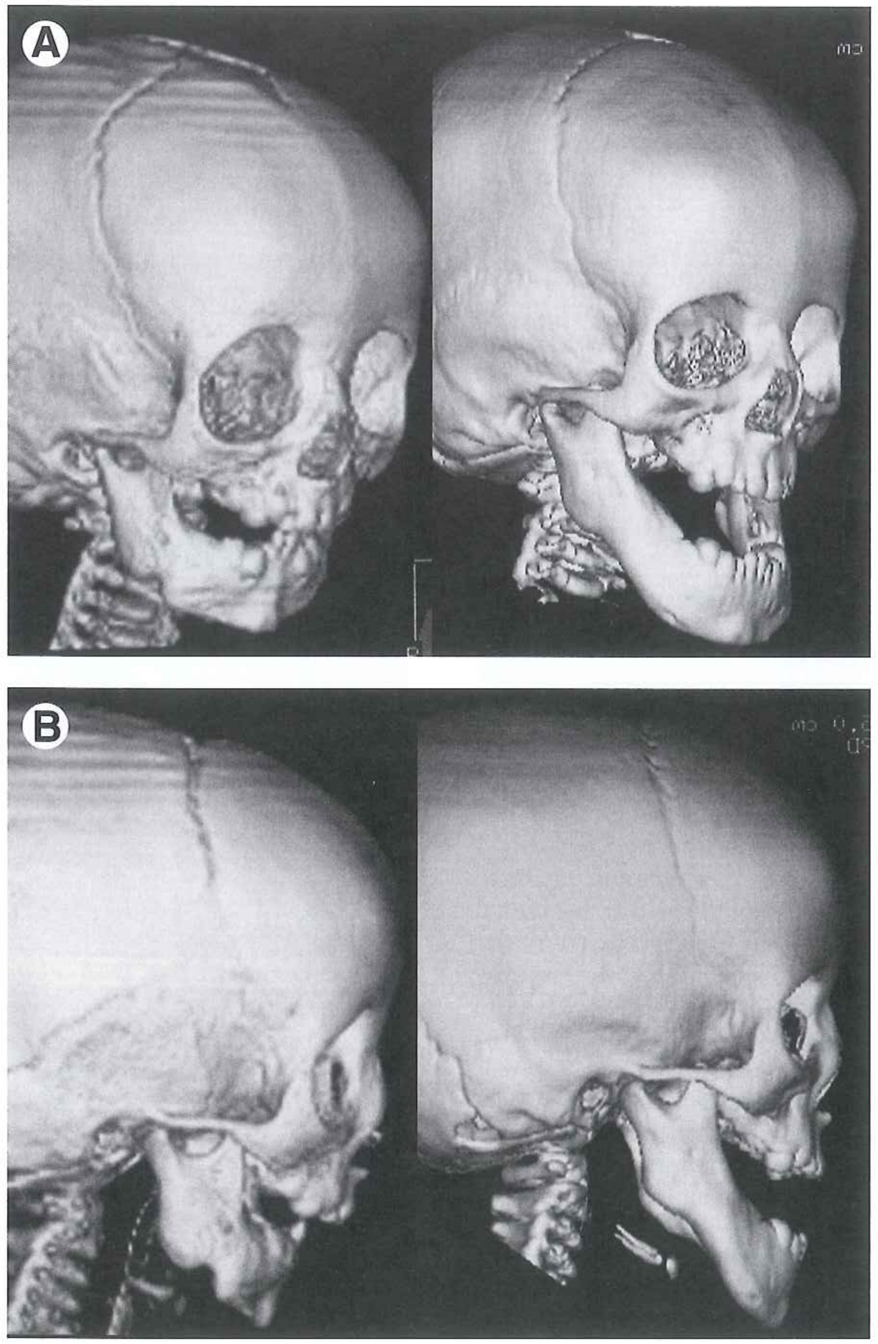

FIGURE 3. Preoperative (*A*) and 4-month postoperative (*B*) computed tomography scan comparisons. Note pin placement marks at mid-ramus and anterior body inferior border.

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per day, and this complication did not occur again after the distraction rate was increased to at least 4 mm per day in the neonatal patient. In 1 patient, the initial distraction devices placed were not long enough to accomplish the necessary magnitude of distraction to achieve improved airway symptoms or a reverse OJ of the occlusion, so the devices were replaced with longer devices. To accomplish this, the carbon fiber rods for the devices that are used during the consolidation period were temporarily placed while the devices were exchanged; the rods were then removed, and distraction progressed as usual. There were 7 cases of hypertrophic scar formation in the area between the transcutaneous pins on the cheeks. In most patients, these became esthetically acceptable over 6 to 9 months following surgery without additional treatment (Fig 7). There were 2 cases of intraoral pin exposure, specifically, the anterior pin

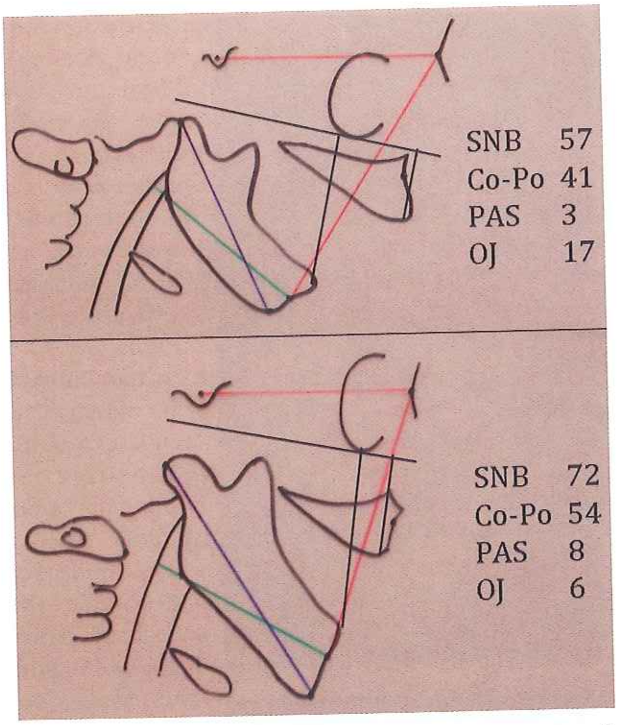

FIGURE 4. Comparison of preoperative and postoperative cephalometric tracings.

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becoming visible in the anterior mandibular buccal vestibule. This likely resulted from placement of the anterior pin too far anteriorly, and resultant movement of the pin through the buccal cortex of the anterior mandible and mucosa. Neither case resulted in loss of distraction movement or in local infection or inflammation. Further, there were no cases of pin site infections either intraorally or extraorally, and there were no cases of development of temporomandibular joint problems, such as hypomobility, disk displacement, or ankylosis. In fact, all patients, and parents, tolerated the distraction procedure well. Most neonates were kept in the hospital during the distraction phase of treatment to be able to control the vector and correct midline discrepancies as necessary because the adjustment of external multivector distraction devices may be confusing for most parents unfamiliar with these devices.

Discussion

The multidisciplinary management of the neonate with airway obstruction remains controversial with a lack of standardization in patient evaluation and management.[21,22] The need for any airway management in the Pierre Robin patient may be less than predicted in the past.[23,24] The usefulness of the tongue-lip adhesion procedure remains variable.[25] Despite the fact that distraction osteogenesis is becoming an accepted treatment modality,[26] the specific techniques of distraction, when performed in the neonatal period, are based on surgeon experience and preference.[27-29] The author's approach to neonatal airway obstruction involves a multidisciplinary management and addressing the feeding issues with an NG tube, Dobhoff, or G-tube or the use of specialized nipples for the cleft palate patient. The airway issues are evaluated with S_pO_2 readings and a measurement of maxillomandibular discrepancy. An objective means should be used to evaluate the airway using radiography and DL. If the DL reveals no obstruction, then a central cause of the problem should be investigated, and the airway

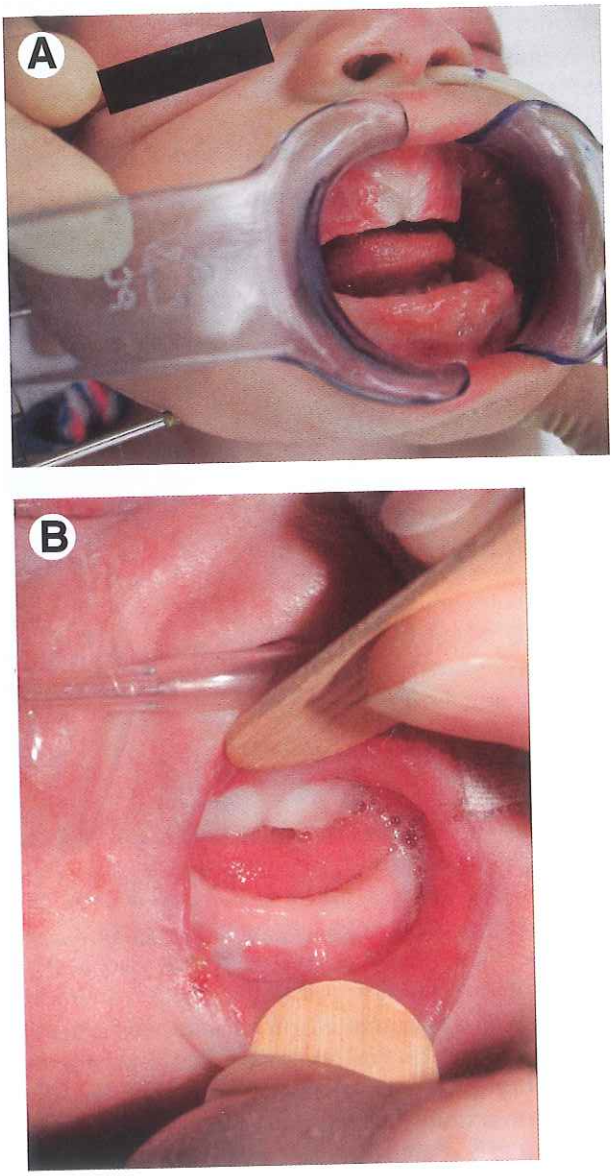

FIGURE 5. Planned overcorrection to a reverse, or negative, overjet relationship.

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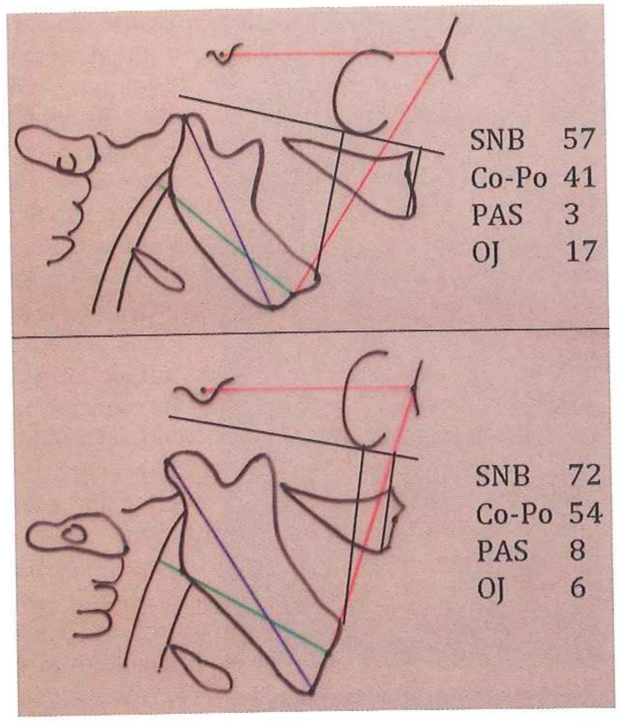

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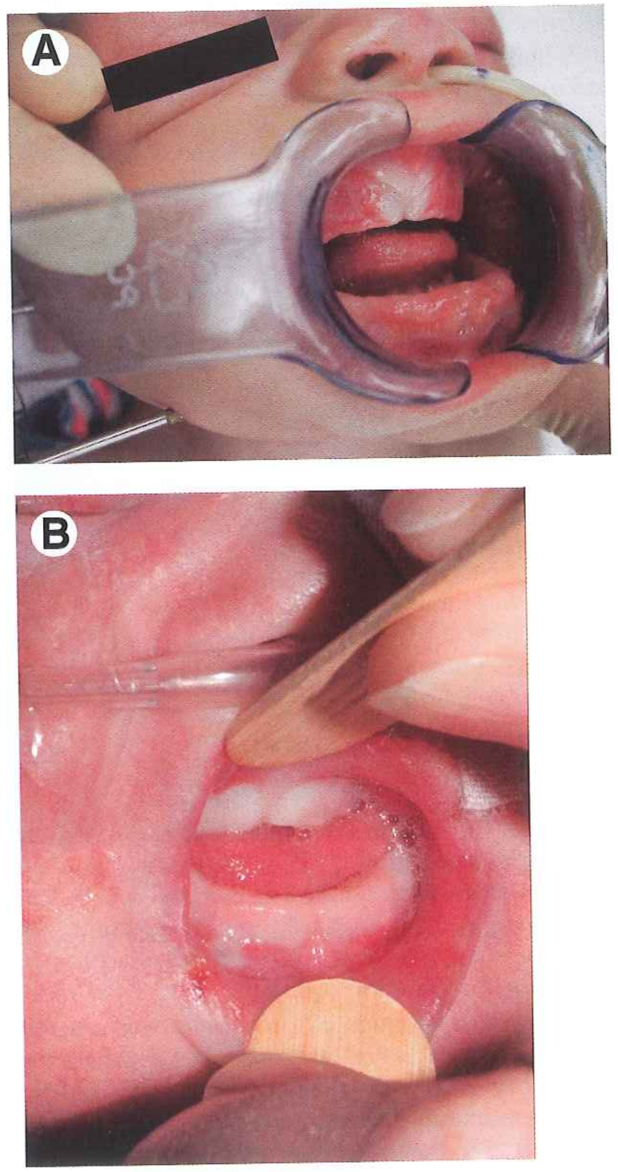

FIGURE 5. Planned overcorrection to a reverse, or negative, overjet relationship.

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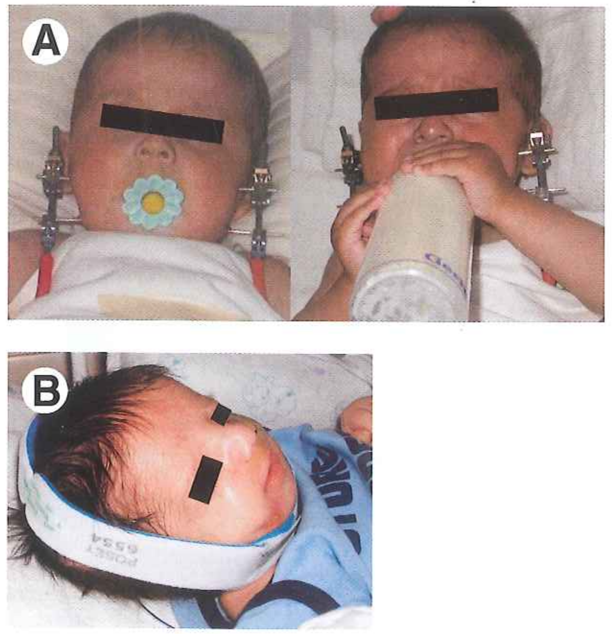

FIGURE 6. *A*, Feeding or nippling during the distraction period to decrease apertognathia. *B*, Use of an elastic head wrap.

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should be controlled with positional maneuvers to maintain S_pO_2. If the DL shows glottic or infraglottic obstruction, then a tracheostomy should be considered, and specialized techniques of laryngoplasty or epiglottoplasty may be used by an experienced surgeon for laryngomalacia or tracheomalacia. Finally, if the DL shows that there is supraglottic obstruction at the tongue base with the posterior pharyngeal wall, then a determination should be made regarding the degree of severity of the symptoms on the basis of clinical parameters, such as cyanosis, frequent apneic monitor triggering with S_pO_2 desaturations, and the magnitude of maxillomandibular discrepancy. For mild cases, head positioning techniques should be used, and tongue-lip adhesion may be considered as a temporary solution before consideration for DO. For moderate cases, airway adjuncts such as a pediatric laryngeal mask airway or nasopharyngeal tube, or tongue-lip adhesion, may be used before consideration for definitive management with DO. For the severe cases, the urgency of the situation, the parent wishes, and the surgeon experience must be evaluated to determine whether tracheostomy or DO should be the first line of therapy.

In addition to those already mentioned, the indications for DO include micrognathia with a maxillomandibular discrepancy, or OJ of more than 8 mm, repetitive upper airway symptoms with chronic low S_pO_2 readings, repeat apnea monitor triggering, labored breathing or cyanosis, poor oral intake, lack of weight gain consistent with age, or abnormal sleep studies (rule out central apnea), and DL or flexible upper airway endoscopy showing obstruction at the tongue base without significant laryngomalacia or tracheomalacia requiring tracheostomy. The patients who should not be considered candidates for DO are those with mild airway compromise controllable with positional head maneuvers; central apnea; and secondary airway lesions, including tracheal webs and malformation of the laryngeal or tracheal cartilages; or a floppy epiglottis.

The benefits of DO are that it treats the underlying problem by lengthening the mandible, elevating the hyoid bone, and advancing the base of the tongue away from the posterior pharyngeal wall to eliminate the obstruction and place the mandible in a forward position for anticipated future normal growth. The issue of growth following DO in the neonatal period has yet to be completely elucidated because the patients who have had this procedure in the new millennium have not reached skeletal maturity for evaluation. Future growth may be variable and unpredictable, however, and therefore, overcorrection of mandibular position from DO has been advocated. Certainly, environmental factors as well as genetic influences may play a role in that syndromic patients with mandibular hypoplasia possess a less inherent growth potential, although most patients may require future orthognathic surgery with mandibular advancement during adolescence.

This specific technique of mandibular DO was chosen because of its relative simplicity and in an attempt to avoid many of the problems associated with DO performed in the neonatal mandible. Of course, the method is technique-sensitive, and inadvertent placement of the transpharyngeal pin may lead to devastating consequences; therefore, constant awareness of

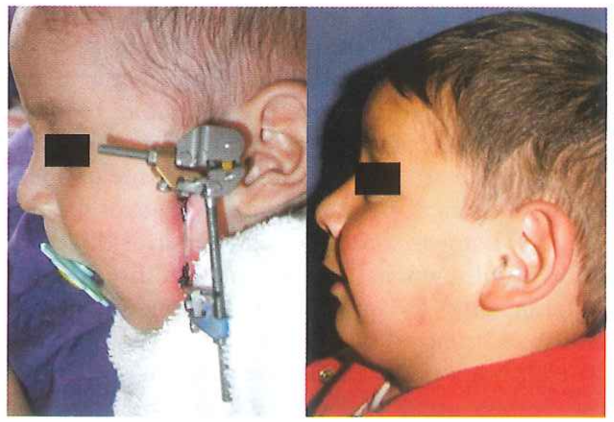

FIGURE 7. Eighteen-month follow-up of distraction osteogenesis–induced facial scar with esthetically pleasing final result.

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the location of the tip of the pin is essential to avoid complications. The technique should not be used by surgeons inexperienced with this specific protocol. The anatomic limitations to device placement include the developing tooth buds and the location of the inferior alveolar nerve in the mandible of young patients. Additionally, the bone is soft and does not allow the firm placement of rigid, or semirigid, fixation devices using monocortical plates and screws. The placement of buried or semiburied distraction devices requiring the use of skin incisions for placement and removal may result in scars and possibly growth disturbance because of fibrosis. It has been shown that the use of pins in one side of the mandible for distraction is not biomechanically stable and that 2.0-mm screws alone cannot withstand the tissue resistance of distraction.[30] The most frequent complication of the creation of an anterior open-bite skeletal relationship can be addressed with physical therapy during distraction as discussed, but the advent of curvilinear distraction devices may allow a more precise positioning of the distal mandibular segment, possibly using predetermined vectors and final mandibular positioning.[31,32]

It should be noted that most major centers that currently perform mandibular distraction osteogenesis for management of the compromised neonatal airway use submerged or semiburied distraction devices placed either transorally or transfacially, and the author (M.M.) has also abandoned the use of the technique described here (used between 2002-2006) in favor of using the newer improved distraction devices. Although the technique described in this article is simple to perform and carries a low risk of morbidity when performed by experienced surgeons, the recent developments in distraction device design, specifically for the pediatric age group, offer many options for distraction. The available devices are low profile and use small diameter and self-drilling screws. The devices may be positioned with a more horizontal vector, parallel to the inferior border of the mandible, to allow a more straightforward mandibular advancement. The "curvilinear" devices address the issue of open-bite development by attempting to control the distraction vector in a forward and superior direction of the distal segment. The advances in three-dimensional imaging and surgical prediction software, as well as customized distraction devices and individualized vector paths enable the surgeon to plan the final segment positions precisely (Fig 8). For these reasons, consideration should be given to the use of the new generation of distraction devices, over the technique described here, although the results presented are certainly acceptable with a low incidence of morbidity and complete resolution of the airway obstruction issues in this group of patients.

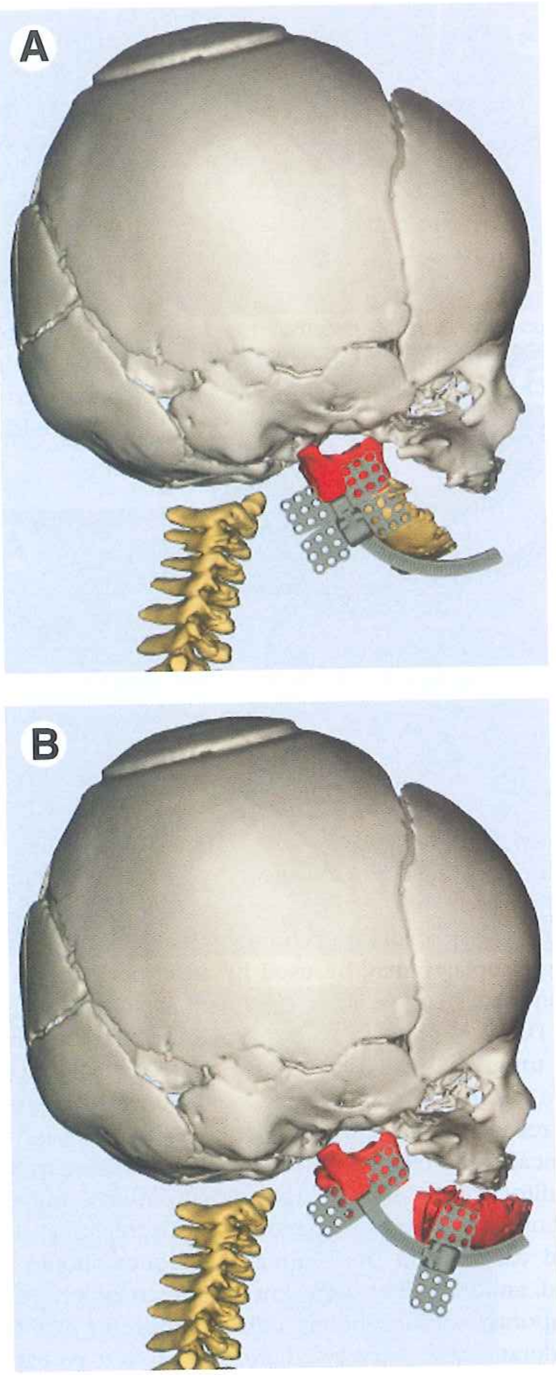

FIGURE 8. Preoperative planning of distraction using a curvilinear device.

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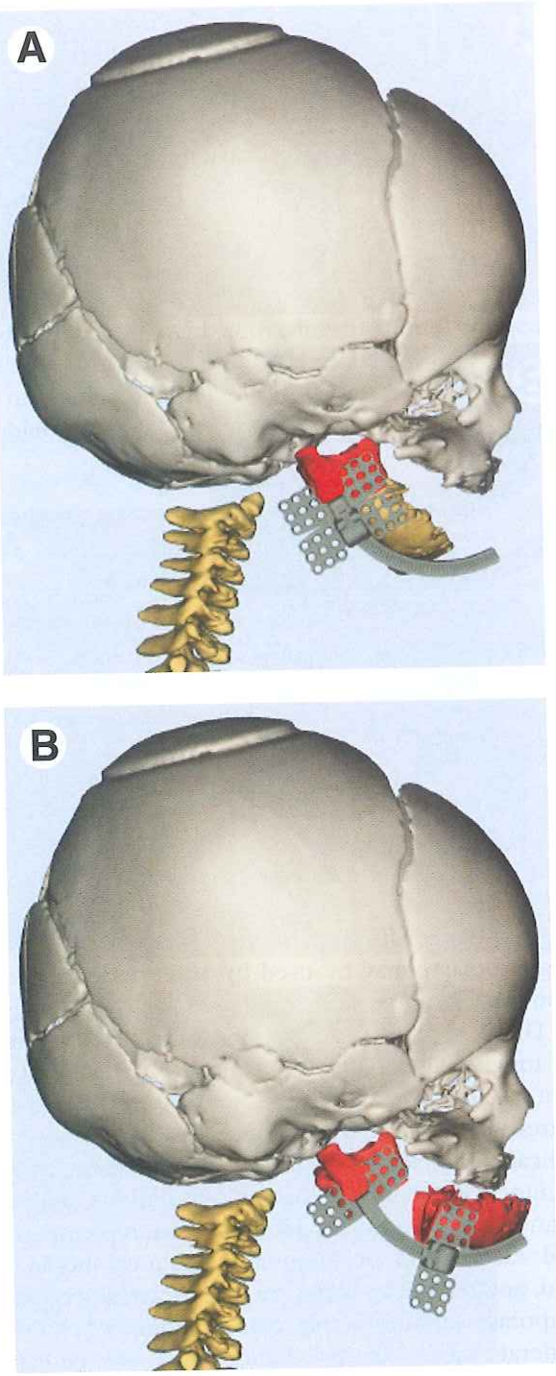

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