

REVIEW ARTICLE

Mandibular Distraction Osteogenesis in the Micrognathic Neonate: A Review for Neonatologists and Pediatricians

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In the past, severe neonatal upper airway obstruction secondary to micrognathia was managed with a tracheostomy. Although effective, tracheostomy can cause many short-term and long-term complications. More recently, mandibular distraction osteogenesis (MDO) has become a well-accepted surgical option in treating micrognathic newborns. Overall, MDO has been reported to be an effective intervention in alleviating the micrognathia-associated airway compromise. Furthermore, it seems to be well tolerated and has supplanted the need for tracheostomy in many patients. Neonatologists and pediatricians commonly care for these children, and therefore an up-to-date clinical narrative review regarding MDO is presented to increase the awareness of this relatively new surgical option.

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1. Introduction

Micrognathia is a congenital condition involving an abnormally undersized mandible. It tends to occur with glossoptosis, and in some cases, U-shaped cleft palate.^{1,2} This constellation of clinical features is now commonly referred to as Pierre Robin sequence (PRS). Craniofacial genetic disorders, such as Stickler syndrome, Treacher Collins

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syndrome, and hemifacial microsomia, can occur in conjunction with PRS. However, PRS is more commonly observed in isolation.²

Micrognathia can lead to upper airway compromise due to posterior tongue collapse and physical obstruction of the oropharyngeal and hypopharyngeal regions. Although most newborns with micrognathia or PRS are either asymptomatic or can be managed with conservative therapy, some patients may have significant airway obstructive and feeding problems.³ Conservative therapy mainly includes close monitoring with prone or lateral decubitus positioning. Other nonsurgical measures involve the use of temporary nasopharyngeal or oropharyngeal airways and the application of nasal continuous positive airway pressure.⁴ Furthermore, as the neonate grows, many will demonstrate catch-up mandibular growth, which occurs at a more rapid rate than other parts of the craniofacial skeleton.²⁻⁴ However, not all patients will respond to these noninvasive interventions.

Significant neonatal upper airway obstruction secondary to micrognathia demands urgent medical attention. Traditionally, tracheostomy has been the gold standard treatment for these patients.⁵ This procedure, however, can cause many short-term and long-term complications.⁶⁻⁸ Therefore, other methods of definitive airway management in this patient population are needed.

Mandibular distraction osteogenesis (MDO) is a relatively new surgical treatment that involves gradually lengthening the mandible to correct the posterior tongue base position to relieve the pharyngeal airway obstruction.⁹ Initially, reports of MDO during the first few days of life were limited, but recent studies have provided more evidence to consider when managing neonates with micrognathia.¹⁰⁻¹⁷

Neonatologists and pediatricians play a crucial role in the perioperative management of hospitalized micrognathic newborns and are often involved in treating the associated airway and feeding problems. This review will discuss the various management issues of micrognathic newborns with emphasis on the safety and efficacy of MDO. The current review will serve to provide up-to-date information regarding MDO for the neonatologist and the pediatrician.

2. Review

A search of the online database of the National Library of Medicine (PubMed) was performed to identify all publications regarding MDO. All studies involving MDO in neonates were included for the narrative review.

2.1. Pierre Robin sequence

The initial insult in PRS is the failed anterior growth of the mandible. The exact cause of this anomaly is unknown but has been hypothesized to be secondary to hyperflexion of the neck in utero.⁹ Some authors also suggest a primary growth disturbance, especially in syndromic PRS cases.⁹ The abnormally small mandible positions the tongue posteriorly and superiorly, which is termed glossoptosis, and can lead to the development of U-shaped cleft palate by preventing the midline fusion of the palatine shelves.⁹

Simultaneously, the retrodisplacement of the tongue can result in oropharyngeal and hypopharyngeal obstruction, leading to varying degrees of respiratory distress.¹⁸ In addition, newborns may struggle with oral feeding, because of problems in coordinating respiration and swallowing in the context of upper airway obstruction and sometimes a cleft palate.²

The diagnosis of micrognathia and PRS is usually made at birth but can be made as early as the second trimester of pregnancy with prenatal high-resolution ultrasonograms. In certain cases, the neonatologist can be called upon during the delivery of prenatally diagnosed children with PRS. However, assessment of the oropharyngeal soft tissue is not easily performed with prenatal ultrasonography and therefore, predictions of which newborn will have airway obstruction is not possible at this time.¹⁵

The most notable feature at birth will be the hypoplastic and retrusive mandible (Figure 1). The lower mandibular alveolus will be significantly posterior to the upper maxillary alveolus. Examination of the oral cavity and oropharynx will usually demonstrate a tongue position that is posterior and superior; some newborns may also have a cleft palate. In patients with syndromic PRS, other craniofacial features may be noted at birth.

2.2. General management of micrognathic newborns

Most neonates with PRS will not require any surgical airway intervention.^{2,19} Many patients will have a mild degree of micrognathia and will subsequently not present with a significant upper airway compromise and/or feeding problems. For those neonates with mild-to-moderate respiratory distress, conservative measures are initially instituted. Specifically, positioning maneuvers with pulse oximetry in an intensive care unit or other monitored units may be sufficient in some children. This is due to the presence of catch-up mandibular growth, which occurs during the first few months of life (mandible grows at a faster rate than other parts of the craniofacial skeleton). However, catch-up growth is not always predictable and can only lead to partial improvement in craniofacial



Figure 1 A photograph demonstrating a hypoplastic and retrusive mandible (micrognathia).

proportions.^{20,21} Furthermore, children with PRS and associated genetic syndromes may not demonstrate any significant catch-up growth.

More invasive interventions include continuous positive airway pressure, nasopharyngeal or oropharyngeal airways, and endotracheal intubation, but they offer only temporary relief of the upper airway obstruction because they are not tolerated for long duration.^{2,19} Mostly, newborns who require prolonged treatment with these measures may require a more definitive surgical intervention.

Currently, three surgical options exist for the micrognathic newborn: tongue–lip adhesion (TLA), tracheostomy, and MDO.

Various techniques of TLAs have been described,²² but they all involve the tongue being surgically fused to the anterior lower lip to hold the tongue in an anterior position. The adhesion is usually reversed with another surgical procedure when the patient is 9–12 months of age. Although TLA has been reported to be successful by some authors, the tethering of the tongue can lead to swallowing difficulties, which may necessitate the placement of nasogastric or gastrostomy feeding tubes.^{2,23–25} Furthermore, the underlying anomaly is not fully addressed, and therefore, many craniofacial centers have abandoned TLA as a viable option.¹⁹ This is also the case at the authors' institution.

Tracheostomy immediately relieves the upper airway obstruction and is still considered the gold standard treatment option by many clinicians. However, the associated mortality rate of 1–4%^{26,27} and the significant morbidity, which includes accidental decannulation, tracheostomy tube obstruction, bleeding, pneumonia, granulation tissue formation, cricoid cartilage injury, swallowing dysfunction, speech and language developmental problems, and late decannulations, make tracheostomy an option fraught with serious limitations.^{5,28–30} Furthermore, children with long-term tracheostomy require nursing care at home and in their educational institutions, along with monitoring and suction equipment.^{28,30} In addition, negative psychosocial consequences for the caregivers and family members of children with tracheostomies have been documented.^{5,30}

2.3. Mandibular distraction osteogenesis

Distraction osteogenesis is a method of producing new bone with a surgical procedure that involves an osteotomy (bony cut) and gradual lengthening of the divided bony segments. Ilizarov³¹ was the first to describe this procedure in long bones on the basis of the “tension–stress” principle. He suggested that the biomechanical factors mediating the process of distraction osteogenesis (gradual lengthening) result in a constant and localized tension and stress, which promotes metabolic activation, angiogenesis, and new bone formation.

The addition of distraction osteogenesis to craniofacial surgery has revolutionized the surgical management of congenital and acquired defects, and the procedure has become widely accepted at several children's hospitals throughout the world. McCarthy and colleagues³² first introduced the use of distraction osteogenesis to lengthen the human mandible for the treatment of hemifacial

microsomia in 1992. Since then, the indications for MDO have rapidly expanded to include airway obstruction in the micrognathic infant (Figures 2 and 3).

Distraction osteogenesis of the mandible relieves the tongue base obstruction by lengthening the mandible. More specifically, because of genioglossus and other muscular attachments of the tongue to the mandible, MDO positions the tongue more anteriorly, thereby reducing the glossoptosis.

The surgical procedure of MDO is composed of three phases: (1) osteotomy and latency, (2) distraction, and (3) consolidation. The first phase involves a surgical procedure where osteotomies are made on the mandible, and distractor devices are placed spanning the proximal and distal segments of the bone to be distracted (Figure 4). The distraction phase soon follows, where the distractor device is activated to gradually lengthen the mandibular bone. The rate of distraction is typically 1–2 mm/day and is performed with a turning device that attaches to an external part of the distractor device. Because the lengthening process is steady and gradual, the overlying soft tissues will stretch to accommodate the newly expanding bony framework. This deliberate process is usually very well tolerated (no analgesia required), and the neonatal patient is able to resume a normal diet during the early postoperative period. Moreover, most patients will demonstrate a notable improvement in their respiratory status after

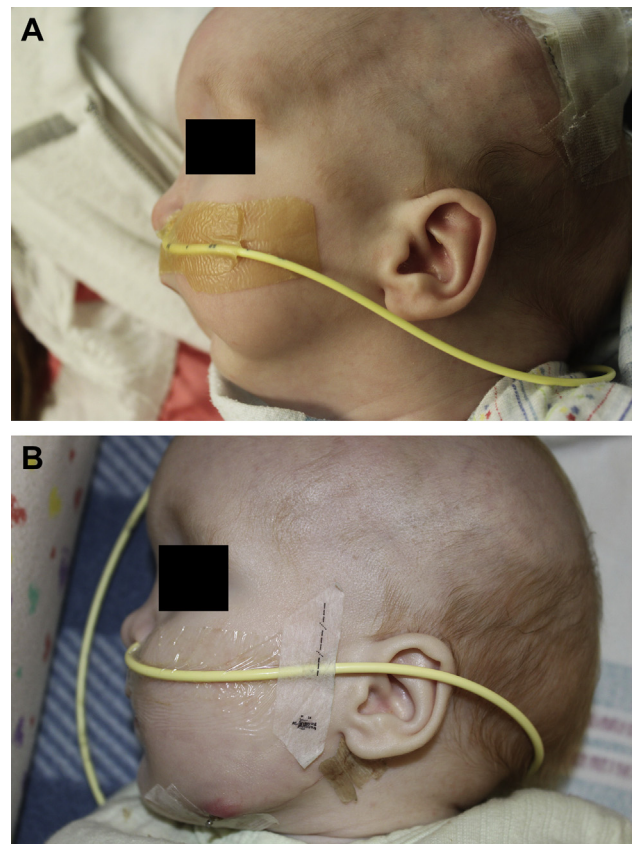


Figure 2 Photographs of a child with micrognathia and airway obstruction (A) before and (B) after mandibular distraction osteogenesis. Note the change in the profile of the lower face.



Figure 3 Photographs of a child with micrognathia and airway obstruction (A) before and (B) after mandibular distraction osteogenesis. Note that the end of the distractor device comes through the skin (B). This part is engaged and turned to gradually advance the mandible.

a few days of distraction (usually 7–14 days), and they can be transferred to a regular hospital ward or even allowed to go home. The final phase is the consolidation phase, which involves healing and solidifying of the newly generated bone. During this phase, which usually takes about 1–2

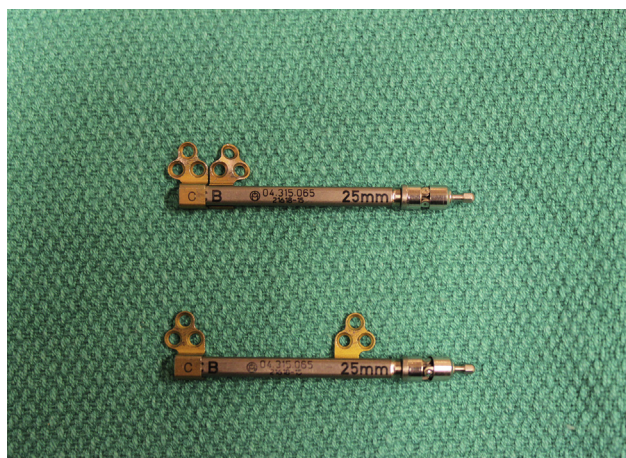


Figure 4 The internal distractor device is placed along the mandible, spanning the osteotomy site. The activator end, which protrudes through the skin, is turned to advance the mandible.

months, the distractor device is left in place to act as a fixation device.

3. Preoperative Investigations

At present, there are no specific indications or guidelines that can be applied when evaluating newborns for MDO. Many surgeons use both subjective and objective measures to assess preoperative candidacy.

3.1. Clinical and laboratory assessments

Preoperative clinical assessments should include comprehensive pulmonary and neurological examinations.^{10,16} Specifically, the degree of respiratory distress and upper airway compromise, oxygen saturation levels, and signs of neurological abnormalities, such as hypotonia, should be documented. For instance, in the presence of generalized hypotonia, MDO should not be performed because the clinical symptoms may not strictly be due to the micrognathia-associated tongue base obstruction, and the patient may still require a tracheostomy or other airway interventions.

Laboratory investigations mainly include blood gases to assess the level of pCO₂. In our institution, a severe obstruction is partially defined as a persistent pCO₂ greater than 50 mmHg, in addition to the clinical findings.

3.2. Feeding and reflux evaluation

Clinical evaluation of the volume, frequency, and quality of successful feeding (bottle or breastfeeding), as well as appropriate weight gain, and the inability to concurrently feed and breath adequately, must be analyzed in detail^{9,14,16} when considering MDO. A diligent assessment of reflux and swallowing dysfunction with pH probe, swallow studies, and feeding evaluations can help determine candidacy, as some children with significant reflux can have poor postoperative outcomes.^{10,11} However, some authors report that feeding and swallowing problems, along with reflux, all tend to improve post-MDO, and therefore reflux should not be considered a contraindication for distraction osteogenesis.^{14,33}

3.3. Imaging workup

Commonly used preoperative imaging studies include radiographs and three-dimensional computed tomography scans (Figure 5), which can readily show the details of the deficient mandible, which allows proper planning of the osteotomies.^{4,10,34} More importantly, the imaging studies can show the location of the tooth buds and the inferior alveolar nerve, which are ideally avoided during surgery.

Newer imaging techniques, such as virtual bronchoscopies, may have a role in evaluating neonates with PRS because distal levels of airway obstruction, such as tracheomalacia, can be ruled out. For such patients with other areas of airway obstruction, relieving the tongue base obstruction with distraction osteogenesis may not obviate the need for a tracheostomy.



Figure 5 Three-dimensional reconstructed computed tomographic image of a neonate with Pierre Robin sequence. Note the hypoplastic mandible.

3.4. Polysomnography

Another objective investigation that can be carried out is polysomnography (PSG) or sleep study. PSG can document the frequency and duration of apneic episodes, as well as the severity of oxygen desaturations.¹⁰ More importantly, PSG will assess for central apneas, which is crucial to rule out for success after MDO.⁴ Several cases of failed distraction procedures have been reported secondary to neurological hypotonia and central hypoventilatory syndromes,^{4,11,16} which may have been noted with a preoperative sleep study.

However, PSG is not always available or easily obtainable at many centers. Furthermore, the obstructive episodes may occur mostly during awake hours. To this end, some have argued that a sleep study is not necessary because acute airway obstruction is not a function of sleep, and the presentation is often worse when patients are more alert and agitated.^{4,10}

3.5. Airway endoscopy

Perhaps the most important preoperative investigation for newborns with micrognathia is airway endoscopy. Awake flexible laryngoscopy in the micrognathic neonate can clearly demonstrate a tongue base obstruction at the level of the oropharynx³⁵; in addition, a jaw thrust maneuver to mimic the airway changes induced by MDO can be performed.³⁵ This maneuver can be a predictor of the potential success of the MDO procedure.

Another important role for airway endoscopy is to assess for other levels of airway obstruction. This involves both awake flexible laryngoscopy to rule out dynamic upper airway obstructive pathologies, such as laryngomalacia,

and formal bronchoscopies to ensure the absence of distal airway lesions, such as tracheomalacia.^{9,11,16} For those newborns with multilevel obstructions, additional airway procedures may be necessary, in addition to MDO.⁴

4. Distraction Osteogenesis and Airway Obstruction

Several case series have demonstrated the effectiveness of MDO in alleviating upper airway obstruction in neonates, infants, and older children with PRS.^{11,14–16,35–39} Most patients were able to avoid tracheostomies and those who already had tracheostomies were able to be decannulated. In addition, a meta-analysis of MDO was performed by Ow and Cheung in 2008.¹⁷ This review studied 178 publications, which yielded 1185 patients. Success in preventing tracheostomy was achieved in 91.3% of neonates and infants, but there were no details regarding the failures.¹⁷ Kolstad and colleagues³⁹ retrospectively examined the effectiveness and complications of MDO in newborns (≤ 35 days old), early infants (36 days to 5 months), and older children (> 5 months). Overall, no significant differences in success rates between the groups were observed, and MDO was successful in 90% (9 of 10) of newborns.

Although there is now little debate about the efficacy of MDO in relieving micrognathia associated airway obstruction, the appropriate age to perform the surgery has not fully been settled. Newborns as young as 5 days old have been successfully managed with MDO,¹⁶ and early surgical intervention seems to be safe and well tolerated. Therefore, the initial concern regarding the small size of the neonatal mandible and the lack of mineralization for MDO operation is not truly valid.

MDO has been reported to be effective in relieving upper airway obstruction in micrognathic neonates, but several authors have reported on the failure of distraction to fully alleviate symptoms of airway compromise to allow avoidance of tracheostomies in some children.^{10,16,40} Specifically, children with syndromic PRS tend to have less successful outcomes, and some have required repeat MDO procedures to achieve full resolution of airway-related symptoms. The low success rate of distraction osteogenesis in children with syndromic micrognathia may be attributed to many different factors such as neurological dysfunction, other levels of airway obstruction, and other medical problems.¹⁰ Subsequently, some authors suggest a more conservative management strategy in this population.^{10,41}

5. Distraction Osteogenesis and Feeding Problems

Neonatal feeding is closely related to the health of the neonatal airway and recent studies on MDO have focused on feeding improvements, as well as airway relief, in children with micrognathia.^{14–16,33} Overall, micrognathic children managed with distraction have improved outcomes in oral feeding and many patients can avoid enteral feeding via nasogastric or gastrostomy tubes.

6. Technical Aspects of Distraction Osteogenesis

6.1. Complications

Management of complications of MDO has improved over time as better devices and equipment became available. Currently, there are two main types of distraction devices (see below) and several different surgical approaches, and each has its own associated complications. Less commonly, they include the following: temporomandibular joint ankylosis, Greenstick fracture, osteomyelitis, tooth bud injury, premature union, malunion, pin or device mobility, device failure, facial nerve paresis/paralysis, cheek abscess, open bite deformity, and dentigerous cyst formation.^{10,15,42} More commonly encountered complications include wound infection, temporary paresthesia, and scarring at external pin sites.^{10,15} Of course, failure to decannulate and requirement of other airway interventions are major complications as already discussed above.

Because MDO is still relatively new, the long-term complications have been poorly documented.^{2,23,24} However, communications at scientific meetings and more recent articles have demonstrated that long-term complications, such as relapse or dental developmental problems, are not common.⁴¹

6.2. Distraction osteogenesis devices

At present, there are two main types of distraction devices: external and internal. External devices (Figure 6) have been in use longer, whereas the internal device (Figure 4) is relatively new. The major advantage of external device is the multidirectional vectors that can be applied during the distraction phase. Subsequently, there is an ability to adjust the direction of advancement after the surgery. Disadvantages include greater risk to marginal mandibular nerve and scars at external pin sites.³⁵ Moreover, owing to the bulkiness of the external devices, the neonatal intensive care unit (NICU) team may not be very



Figure 6 An external mandibular distractor device (KLS Martin, LP, Jacksonville, FL, USA).

comfortable with managing these newborns in the early postoperative period. More specifically, if inadvertent extubation occurred during the early phase of mandibular advancement, the NICU team may be uncomfortable with reintubation.

Internal devices are becoming more popular as there is no cumbersome external device to deal with during the distraction and consolidation periods, and there is no risk of pin-associated scar formation or infection. The major disadvantage of the internal distraction device is the unidirectional or linear vector of movement, which requires meticulous planning of osteotomies and distractor placement in the operating room.

6.3. Extubation and decannulation of tracheostomy

Removal of the endotracheal tube after the operation can occur at any time when the airway caliber has improved. This can occur during the distraction phase or at the start of the consolidation phase.

Decannulation for previously tracheostomized children should involve comprehensive airway endoscopy, including flexible laryngoscopy and bronchoscopy. The removal of tracheostomy typically occurs at the time of the distractor removal at the end of the consolidation phase or approximately 1–2 months after distractor removal.⁴

6.4. Cost-effectiveness of distraction osteogenesis

As mentioned above, tracheostomy is associated with complications, long after the procedure has been performed.^{6–8} After tracheostomy, a prolonged hospital stay is typical, because the tracheostomy tube has to be changed on multiple occasions and the caregivers require adequate time to be educated on tracheostomy-associated care. In addition, there is a great long-term cost related to tracheostomy care (see above).

Distraction osteogenesis also bears high costs owing to the operative equipment but the duration of stay in the NICU and hospital tends to be less when compared to children who undergo tracheostomy.⁴³ Moreover, there are typically no long-term costs associated with patients undergoing MDO.

Two cost studies comparing tracheostomy versus MDO both demonstrated that MDO seems to be more cost effective.^{43,44} This is in keeping with the fact that MDO patients are usually discharged home earlier.

6.5. Future direction of distraction osteogenesis

The future distraction of MDO lies in new technological developments and refinements of the procedure itself. Three-dimensional surgical planning with novel imaging software and resorbable distraction devices that have sufficient strength are under development, which should make the operation more efficient and precise.

Moreover, further understanding of the biochemical and molecular mechanisms involved in MDO will lead to the enhancement and hastening of the bone healing consolidation phase.^{45–47}

7. Conclusion

Although most children with PRS can be managed with conservative measures, MDO in the neonate with micrognathia can be an effective and well-tolerated treatment option to relieve the upper airway obstruction. It should be considered as an acceptable alternative to tracheostomy in selected patients.

For managing newborns with PRS, a multidisciplinary approach involving a neonatal intensivist or a pediatrician with experience in neonatal respiratory medicine, a pediatric anesthesiologist, and a craniofacial surgeon, along with other allied health professionals, is preferred.

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