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Feeding Complications in a Six-Week-Old Infant Secondary to Distraction Osteogenesis for Airway Obstruction: A Case Report

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Abnormal breathing during sleep may frequently occur in early childhood, but it is usually unnoticed because of unperceived medical or physiologic sequelae.¹ Infants and children with neuromuscular disease exhibit a higher incidence of obstructive sleep apnea secondary to hypotonicity of the pharyngeal musculature that allows collapse of the airway.^{2,3} In addition, the potential for airway obstruction is further increased by supine positioning, neck flexion, and increased secretions during sleep.²

Initial management of obstructive sleep apnea in children has traditionally included pharmacologic treatment, the use of nasopharyngeal airway devices, oxygen supplementation, and positive airway pressure.^{3,4} When these fail, surgical procedures such as tonsillectomy, adenoidectomy, uvulopalatopharyngoplasty, or tracheostomy may be required to correct or bypass the abnormal anatomic features contributing to obstruction of the airway. Recent literature has suggested that facial skeletal advancement using distraction osteogenesis might be beneficial in those

children with obstructive sleep apnea secondary to midface hypoplasia or retromicrognathia and lack of tongue support.^{2,5,6}

Early distraction procedures used extraoral devices that caused facial scarring. More recently, intraoral appliances have been developed that can avoid this complication as well as the potential for facial nerve damage. To our knowledge, the literature contains no reports of the effects that these intraoral devices, or the resulting anatomic changes, may have on the ability of infants and children to feed during and after the distraction process. We report a case of a 2-month-old infant who underwent successful distraction osteogenesis for treatment of airway obstruction due to retromicrognathia and glossoposis. However, the child had difficulty in feeding during the distraction procedure and for a period after removal of the devices. The case is discussed with emphasis on feeding ability and growth curves.

Report of Case

A 6-week-old, nonsyndromic black infant girl was seen by her pediatrician 3 times before hospital admission with a problem of upper airway congestion and stridor since birth. No cyanosis was noted. The infant was treated with Beconase (Glaxo-Wellcome, Research Triangle Park, NC), but continued to demonstrate stridor and suprasternal retraction and, when placed in a supine position, her upper airway obstruction worsened. She was otherwise healthy.

The patient was subsequently admitted to the pediatric service for evaluation and treatment. Her admitting vital signs were blood pressure, 98/57; pulse 156 beats/min; respirations, 60 breaths/min; and temperature, 99.9°F. Hematologic evaluation was performed, which showed a hematocrit and hemoglobin within normal limits and an elevated white blood cell count (WBC) of 29.8/μL. Her preadmission

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growth evaluation placed her in the 50th percentile for height (57 cm) and in the 25th percentile for weight (4.09 kg). It was noted that she bottle-fed well.

The infant was subsequently started on nebulized racemic epinephrine, neosynephrine, and intravenous Ancel, 120 mg IV every 8 hours, for the respiratory infection. Continuous oxygen supplementation helped to achieve oxygen saturations above 93%. However, she continued to have intermittent episodes of desaturation below 70%, requiring transfer to the pediatric intensive care unit. A bronchoscopy was performed, which showed mild laryngotracheal malacia. The patient was started on cisapride after an esophogram that showed dilation due to chronic reflux.

The pediatric oral and maxillofacial surgery service was consulted on hospital day 10. Examination showed moderate micrognathia, glossoprosis, continued stridor, and oxygen desaturation. A nasopharyngeal tube was placed with minimal improvement. Treatment choices, including tracheostomy and mandibular advancement using distraction osteogenesis, were discussed in detail with the parents. They chose mandibular advancement using the distraction osteogenesis procedure.

The infant was taken to the operating room and, under general anesthesia, bilateral posterior mandibular buccal corticotomies were made. The intraoral distraction devices (KLS Martin L.P., Jacksonville, FL) were placed bilaterally (Fig 1) and the lingual plates were then fractured, completing the osteotomies. The devices were fixed with four 1.5-mm self-drilling screws at the inferior border of the mandible. Primary closure of the soft tissues over the body of the distractors was obtained by advancing the buccal mucosa. The activation arms exited the oral cavity at the commissures, but did not place tension on the lips or interfere with the occlusion (Fig 2).

The patient tolerated the procedure well. She remained intubated and was transferred to the pediatric intensive care unit. Distraction of 1.0 mm per day (0.5 mm $\times$ 2) was initiated on the third postoperative day. Orogastric feeding was used for nutritional support while the child was intubated. The patient was extubated without complication on the fifth postoperative day (distraction day 3), and oral feedings were started. She was noted to have difficulty forming a seal around her bottle. A speech therapist evaluated the patient and noted an intact swallowing mechanism. A cleft palate nipple was recommended, which she tolerated well, taking approximately 90 mL (3 oz) of formula every 3 hours.

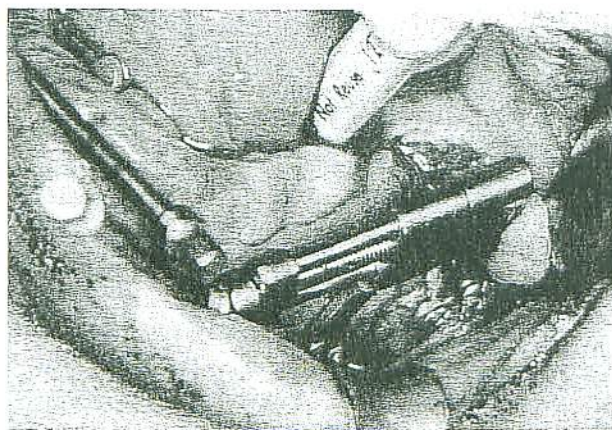


FIGURE 1. Intraoral photograph showing the osteotomy between the device footplates.

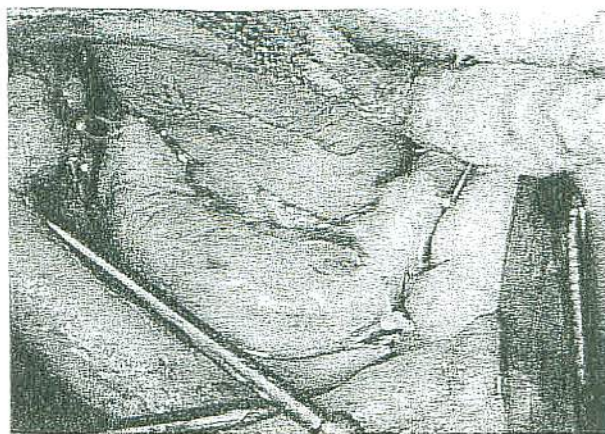


FIGURE 2. Intraoral photograph after wound closure.

Progressive improvement in her respiratory status was evident during distraction. Initially she required supplemental oxygen by nasal cannula; however, by distraction day 5 the patient was able to maintain an oxygen saturation level of 98% to 100% on room air. Active distraction continued for another 1.5 mm, for a total advancement of 6.5 mm (Figs 3 and 4). The additional 1.5-mm advancement was done to allow for potential relapse. Continuous oxygen saturation levels of 98% or greater were maintained and the infant was discharged on the ninth postoperative day.

The patient was readmitted to the pediatric service approximately 4 weeks after discharge for an upper respiratory tract infection, at which time there was no evidence of obstructed breathing. Additionally, decreased subcutaneous fat and a weight of 3.97 kg were noted, placing her below the 5th percentile for weight. Another speech therapy evaluation was performed, and it was observed that the infant did not exhibit a rooting reflex. In addition, she chronically postured her mouth in an open position. Importantly, she exhibited a weak, inconsistent ability to suck. The impression of the speech therapy service was that the child had a dysfunctional and disorganized feeding mechanism.

During this admission (the fifth postoperative week), the

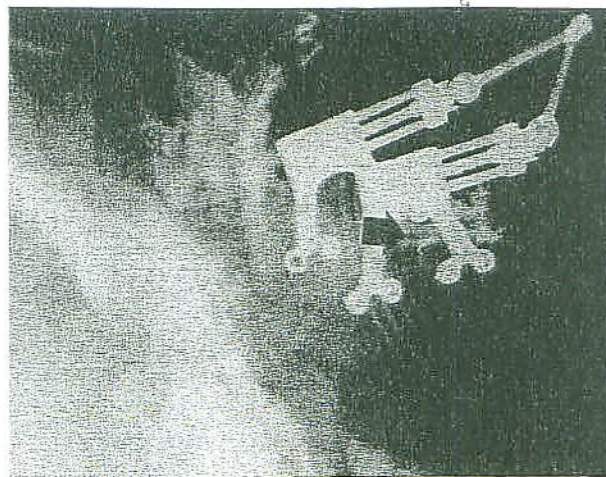


FIGURE 3. Predistraction radiograph showing the appliance in place. The arrow points to the osteotomy site.

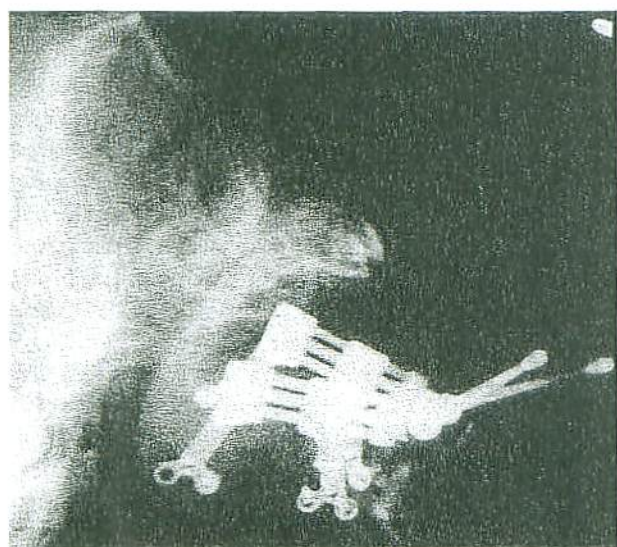


FIGURE 4. Postdistraction radiograph with the appliance in place.

infant was taken to the operating room by the pediatric oral and maxillofacial surgery service, and the distractors were removed without difficulty. Before discharge, the child was able to use a regular nipple with assistance, but she continued to exhibit a weak sucking mechanism.

Three weeks after removal of the distractors, the parents reported that the infant was using a regular nipple without difficulty and exhibited a stronger sucking mechanism. Her weight had increased to 5.40 kg (a gain of 1.40 kg over a 3-week period), which placed her in the 25th percentile range.

Discussion

The treatment of selected cases of upper airway obstruction by mandibular distraction osteogenesis is supported by the recent literature.^{3,4,6} Cohen and colleagues discussed in detail the relationship between obstructive sleep apnea and children with craniofacial disorders, as well as the use of distraction osteogenesis to avoid tracheostomies in these children.^{2,4,5} Guyette et al⁶ reported on 2 patients who experienced changes in their articulation and velopharyngeal function after unilateral mandibular distraction.⁶ Immediately after the completion of distraction, in both of these cases it was noted that articulation skills had declined and velopharyngeal inadequacy was evident. However, in both instances, the velopharyngeal inadequacy was temporary, resolving between 1 and 8 months.

Cohen et al performed distraction in children as young as 14 weeks of age using external devices.⁴ In the current case, intraoral devices were placed on the mandible of an 8-week-old infant who required hospitalization because of desaturation while sleeping in spite of the use of supplemental oxygen and a nasopharyngeal tube. Her immediate postoperative course was uneventful, and she was weaned off supplemental

oxygen 48 hours after extubation. An unexpected sequelae to the procedure was her inability to feed postoperatively, resulting in a significant lack of weight gain.

Normally, an infant is born with an efficient sucking mechanism. The tongue and jaw play a primary role in this process. The lips move secondarily with the jaw without contributing to sucking, but they do provide an efficient seal to reduce liquid loss. A 1-month-old infant will take between 2 and 6 ounces of liquid per feeding. The child will swallow using a suckle-swallow pattern, and the tongue may protrude slightly through the lips in an extension-retraction pattern. The rooting reaction aids in the search for liquids. Between 1 and 3 months, the child will lose that physiologic flexion, and the backward/forward tongue movements of a suckle emerge. During this transition period, there may be some fluid loss because of a sucking pattern that is less tight than it was at birth. At 3 months, infants will take between 7 and 8 ounces of liquid per feeding and use only suckling and sucking patterns. By 4 months, a child should recognize a bottle or spoon and will suckle in anticipation. The child may cough and choke more frequently while learning to swallow as the neck elongates.^{7,9}

Significant anatomic differences exist between the oropharynx of a newborn and an adult. The mandible of a newborn is small and retracted, resulting in a small oral cavity and the impression of an oversized tongue. The tongue fills the entire oral cavity and touches the floor and the roof of the mouth simultaneously. The amount and direction of tongue movement is limited in newborns and young infants. Because the tongue at rest is in direct contact with the palate, there is no additional space to support up and down tongue movements. This is one reason why the direction of early tongue movement in the suckle is backward and forward.^{7,9}

Newborns are obligate nose breathers because the mouth is filled with the tongue and the soft palate and epiglottis are in contact. The infant's epiglottis and soft palate are in direct approximation until the child is 3 to 4 months old. This allows the infant to swallow food and pass it laterally onto the outer edges of the epiglottis and then into the pharynx and esophagus, protecting the airway at a time when the infant is neurologically immature.^{7,9}

The early stage of continuous tongue-oral cavity contact is essential to the acquisition of sensorimotor information. As the tongue gains more space in which to maneuver, it simultaneously gains greater neurologic control. The increase in up-and-down movement of the tongue coincides with a greater capacity for control of the lip valve, thus improving negative intraoral pressure. Developing structural changes in infant anatomy allow for more mature feeding pat-

terns, but also may complicate the oral-motor development for neurologically impaired infants.^{7,9}

This patient exhibited initial tongue retraction and micrognathia. Her tongue was positioned so that the tip rested in the posterior third of the oral cavity. This in itself can create obstruction of the pharyngeal airway and cause a noisy, stressed breathing pattern. In addition, low or high neuromuscular tone can create exaggerated early extensor movements, and tongue retraction can result.⁸ The combination of tongue retraction, micrognathia, and mild tracheomalacia contributed to this patient's obstructed breathing pattern. Mandibular distraction osteogenesis was successful in repositioning the osseous and soft tissue structures, thus enlarging the oropharynx and giving the infant the opportunity to neurologically mature and develop better control of her upper airway.

The intraoral appliances directly prevented the infant from forming a seal with her lips during the 5 weeks they were in place. Initially, after appliance removal, the infant showed a lack of rooting, an open mouth posture, occasional lip closure, and a weak, inconsistent ability to suck. She continued to require a special cleft palate bottle, showed saliva pooling in the back of the mouth, and had several episodes of coughing during feeding. During the following week, her open mouth posture decreased to a more appropriate, relaxed position, and the amount of liquid taken at each feeding increased. She gained over 1.4 kg (3 lbs) in the 3 weeks after discharge, and her weight was approximately 5.40 kg.

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Pneumoparotid in Childhood: Report of Two Cases

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When considering the use of mandibular distraction for an infant who exhibits signs of neurologic immaturity, such as tongue retraction, obstructed breathing, and uncoordinated feeding, one should also be prepared for a transient decrease in the ability to feed. The use of feeding appliances, such as the cleft palate nipple, may be required while the distractors are in place as well as after their removal, as the child adapts neurologically to their rapid anatomic changes.

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Enlargement of the parotid gland due to air insufflation has been called by such names as *pneumoparotitis*,¹ *pneumoparotiditis*,² *pneumosaladenitis*,³ *pneumatocoele glandulae parotis*,⁴ *wind parotitis*,⁵ and *anesthesia mumps*,⁶ but the most appropriate term is *pneumoparotid*.⁷ Goguen et al⁸ proposed a division into 2 categories: isolated acute events (transient) and recurrent injuries. Acute pneumoparotid has been associated with dental procedures in which high-speed air turbine handpieces⁹ and air-powder prophylactic cleaning units,¹⁰ which increase the normal intraoral pressure (2 to 3 mm Hg) up to 20 times, are used near the molar teeth. Likewise, *anesthesia*