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Clinical Paper
Cleft Lip and Palate

A new way to anchor the external device in mandibular distraction: three case reports with a Pierre Robin sequence

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Abstract. Pierre Robin sequence is a pathology derived from alteration in the first and second branchial arch. Patients have breathing problems due to micrognathia and glossoptosis, causing severe upper airway obstruction. One surgical treatment is distraction osteogenesis. Three patients with Pierre Robin sequence (case 1, 3 months old; cases 2 and 3, 1 month old) with severe upper airway obstruction requiring mechanical ventilator assistance, underwent mandibular distraction osteogenesis prematurely with a new anchoring system, thus avoiding tracheostomy and its consequences. An intraoral approach was used to avoid scarring. A new anchoring device with transfixing Kirschner wire in the proximal (mandibular ramus) and distal segment (chin zone) was used. This diminishes the risk of distractor device displacement, guaranteeing optimal stability. A more anterior installation reduces the risk of damaging tooth buds in the mandibular body and the inferior alveolar nerve. The more anterior the fixation, the more horizontal the distraction vector becomes. The position and stability of the device are crucial. In these three patients the placement of two transfixing Kirschner wires using an intraoral approach showed good results and stability during the period of distraction and consolidation, with optimal results on the upper airway, avoiding tracheostomy.

Keywords: distraction osteogenesis; Pierre Robin; micrognathia; upper airway obstruction.

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The Pierre Robin sequence is one of the pathologies caused by alterations in the first and second branchial arch, characterized by mandibular hypoplasia, resulting in a cleft palate and glossoptosis[8]. A cleft palate makes sucking and swallowing difficult, allowing fluids easy access into the larynx and glossoptosis. It contributes to respiratory alteration. The poor development of the mandible results in an inadequate space for the tongue to descend. The vertical position of the tongue is the main factor that obstructs the horizontal positioning of the palate, causing a cleft palate.

There are several physiological alterations derived from this sequence. The most serious and complex alteration is upper airway compromise. The upper airway obstruction ranges from simple chronic airway limitation and episodic apneas, to severe obstruction of ventilation causing acute respiratory insufficiency. Swallowing alterations can make oral nutrition difficult, which frequently leads to malnutrition. Gastroesophageal reflux, secondary to the chronic limitation of the upper airway, contributes to the nutritional problem, often requiring the installation of a nasogastric probe or even a gastrectomy[6].

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In the 1990s, bone distraction began to be practiced in the maxillofacial area. Mccarthy et al.[4] and Molina and Ortiz-monasterio[7] made preliminary efforts to achieve the growth of bone and soft surrounding tissue.

The purpose of this report is to present a series of three cases of newborn patients carrying the Pierre Robin sequence, with severe obstruction of the upper airway, who underwent mandibular distraction osteogenesis via the intraoral approach, using two transfixing Kirschner wires, proximal and distal. In all three cases, mandibular distraction osteogenesis avoided tracheostomy and its consequences.

Patients and methods

Three newborn patients diagnosed with Pierre Robin sequence with severe episodes of obstructive apnea, with altered polysomnography, underwent bilateral mandibular osteodistraction via the intraoral approach with the placement of two external fixation devices using two transfixing Kirschner wires. None of the patients required tracheostomy before or after surgical treatment.

Case 1 was a 3-month-old child with Pierre Robin sequence who experienced repeated episodes of obstructive apneas, which were treated initially with oxygen therapy. After a few days, the respiratory obstruction deteriorated, causing the patient to be admitted to the intensive care unit, where mechanical ventilation was required. A complete examination and study of the patient were carried out, which confirmed the diagnosis of Pierre Robin sequence, permanent and severe bronchial obstructive syndrome, oxygen dependency, swallowing disruption, gastroesophageal reflux and chronic malnutrition.

An osteogenic, bilateral mandibular distraction was proposed to correct the micrognathia and stimulate the suprahyoid muscles to gradually enlarge the upper airway, the movement of which corrects the position of the tongue in the oral cavity.

Case 2 was a 1-month-old child with Pierre Robin sequence who was admitted to the intensive care unit, where mechanical ventilation was required. An osteogenic, bilateral mandibular distraction was proposed with two external fixation devices, with two transfixing Kirschner wires.

Case 3 was a 1-month-old child with Pierre Robin sequence who was admitted to the intermediate care unit, due to an obstructive sleep apnea, low weight and deglutition problems. An osteogenic, bilateral mandibular distraction was proposed with two external fixation devices, with two transfixing Kirschner wires.

Surgical technique

Under general anaesthesia and orotracheal intubation, an intraoral approach on both the mandibular body and the ramus was made. Osteotomies in both mandibular rami behind each angle were performed very carefully to avoid damaging the dental organs which were in intra-osseous evolution.

The osteotomized area was delimited using superficial corticotomy throughout all the external face of the mandible. Afterwards, osteotomy of the alveolar and basilar edges was performed, making a superficial vestibule cut, whilst being careful to preserve the inferior alveolar nerve. A transfixing Kirschner wire was installed through both proximal segments in the ramus area (right and left), being careful to keep it completely horizontal. Another transfixing Kirschner wire was installed and anchored from side to side of the symphysis, parallel to the previous one (Fig. 1). Before the second transfixing Kirschner wire was installed, the skin between the wires was pinched to minimize the scar after the distraction. The Molina external distractor (Wells Johnson Co.) was installed on each mandibular side. Both sides were activated until complete osteotomy was reached, returning the mandibular sides to their original position.

Distraction protocol

Three days later activation was initiated with a 1.0 mm magnitude every 12 h for 3 days. Traction continued with 0.5 mm every 12 h during the subsequent days depending on the amount of distraction desired.

After the activation was complete, contention was performed for 4 weeks, keeping the distractor device in a static position. Afterwards the distractor device was removed without requiring general anaesthesia.

During osteogenic distraction, suction was stimulated, and mandibular lateral movements were performed with the guidance of an operator (nurse), to stimulate growth and avoid ankylosis due to the pressure over the temporomandibular joint[10].

Results

The results at the end of the activation period were 20, 25 and 23 mm, respectively, for each case. The patients achieved normal respiratory physiology, with normal polysomnography, complete horizontal positioning of the tongue, and a larger upper airway area. Gastroesophageal reflux was cured, confirmed with pH measurements. Adequate oral nutrition was achieved with the help of suction stimulation through a pacifier and feeding bottle. The balance of the maxillomandibular relationship was corrected and proper chin projection, soft tissue coverage, and adequate positioning of the alveolar bones were achieved immediately post-distraction. There was no skeletal open bite in any patient (Figs. 2–4). The Kirschner wires did not move and were not released during the distraction in any of the cases, showing great stability.

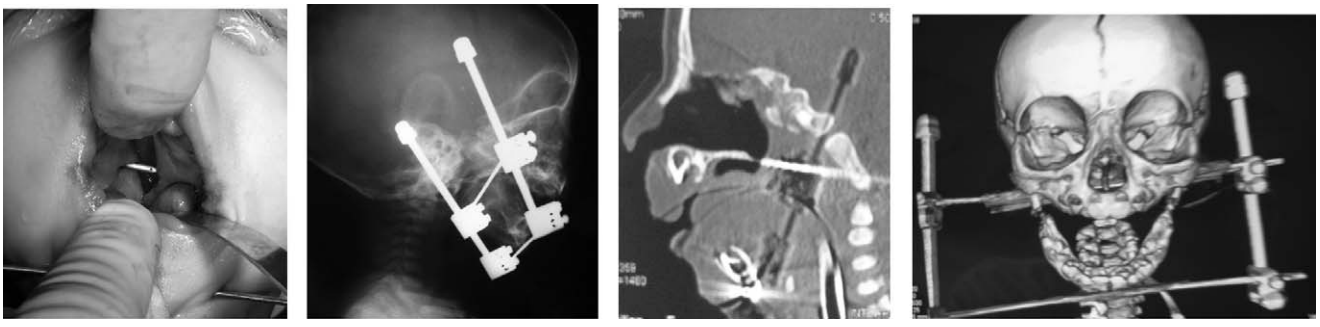

Fig. 1. Transfixing Kirschner wires: (a) intraoperative view; (b and c) radiographic views and (d) computer tomography scan.

Fig. 2. Three-month-old child. (a) Preoperative profile and (b) postoperative profile at 4-year follow up. Radiographs: (c) preoperative and (d) postoperative after 20 mm of distraction.

Fig. 3. (a) One-month-old child with Pierre Robin sequence; (b) during consolidation phase; (c) at 3-year follow up after 23 mm of distraction.

Fig. 4. (a) One-month-old child with Pierre Robin sequence; (b) after 25 mm of distraction, at 2-year follow up.

In all cases, the growth and development of the jaw is completely normal after 2, 3 and 4 years of follow-up. The dental buds were preserved and they are in adequate positions. The alveolar nerve is normal. Mandibular mobility is normal. There are no restrictions to mouth opening.

Discussion

Upper airway management with the Pierre Robin sequence has always been a hazard owing to the severe structural deficiency of the lowest third of the face. When planning therapeutic strategies, etiologic solutions that offer favourable clinical outcomes should be used. Osteogenic distraction is a relatively new technique used to achieve mandibular enlargement. Based on bone physiology and biomechanical concepts, an initial osteotomy and posterior gradual traction provide a good solution for patients with alterations of the upper airway due to mandibular growth and less developed alterations.

McCarthy et al.[4] and Molina and Ortiz-Monasterio[7] propose a sub-mandibular approach, and an osteotomy in the angle area of the mandibular ramus, installing pins close to the line of fracture. The present authors perform an intraoral approach to avoid scarring the skin, and to achieve minor soft tissue trauma with less possibility of facial nerve damage[9]. The osteotomy is done behind the last tooth bud in an oblique form, but the authors prefer to instal a transfixing Kirshner wire in the proximal segment and in the mental zone. This is done for three reasons: it significantly diminishes the risk of displacement of the distractor device,

guaranteeing optimal stability; the more forward the wire is fixed, the lower the risk of damaging tooth buds or the alveolar nerve in the mandibular body; and the more anterior the device is fixed, the more horizontal becomes the distraction vector.

Molina and Ortiz-monasterio[7] use pin installation, first proximally and then distally. When using a transfixing Kirschner wire in the proximal and chin zone, the present authors suggest the same sequence to achieve the desired parallelism between both wires easily. Gifford et al., in a report of eight cases of neonates with mandibular hypoplasia, comparing mandibular distraction osteogenesis Kirschner wires versus screw fixation devices, established that titanium screws offer greater stability than Kirchner devices in the jaws of infants[3]. The authors' technique offers great stability with two external fixation devices, with two transfixing Kirschner wires, at the proximal and distal area.

The authors started activation of the distractor on the third day after surgery, with 1 mm every 12 h for the first 3 days, continuing with 0.5 mm every 12 h for the next days. Mccarthy et al.[5], when presenting their 10-year experience in osteogenic distraction, noted that the latency period before initiating activation should be 5 days, keeping a rhythm every 12 h during traction. They emphasized that in those patients who were too young, activation should be of 1.5 mm a day, due to their great metabolic potential and the risk of premature consolidation. Molina and Ortiz-monasterio[7] recommend 5 days as the latency period, after which they continue with distraction of 1 mm every 24 h. Denny and Kalantarian[1], when presenting a series of six patients with obstructive airway disorders, who had undergone osteogenic distraction, started activation the day after the intervention, with a rhythm of 2 mm every day, for the first 3 days. Sadakah et al. reported seven cases of bilateral intraoral distraction osteogenesis in patients with severe mandibular hypoplasia, establishing a latent period of 3 days in their distraction protocol, and active distraction at a rate of 0.5 mm every 12 h for 17–24 days[9]. After analyzing the different reports, the present authors decided to wait 3 days to allow a clot to form in the osteotomized zone. Activation was started with 1 mm every 12 h to allow a greater initial advance, aiming at quickly dealing with the obstruction of the upper airway. Distraction was carried out every 12 h rather than every 24 h, because this diminishes the separation magnitude of the two segments during each activation, reducing pain in the patient.

Consolidation time is also a controversial issue. Denny and Kalantarian[1] propose 2 days of contention for every day of distraction. Molina and Ortiz-monasterio suggest stopping contention when the mature bone can be visualized radiographically, after approximately 6–8 weeks[7]. Felemovicius and Ortiz-monasterio[2] propose different time of consolidations depending on the age of the patient, based on osseous metabolism demonstrated by scintigraphy. Sadakah et al. established a consolidation period of 4 weeks, with highly satisfactory results[9]. In patients under 1 year, only 4 weeks of contention phase are necessary.

In conclusion, mandibular distraction is an adequate surgical treatment for patients with the Pierre Robin sequence with obstruction of the upper airway. Mandibular distraction is an alterative to tracheostomy in these patients. The anchorage position and stability obtained with transfixing Kirschner wires fixed in the mandibular ramus and symphysis are crucial for obtaining excellent results in mandibular osteogenic distraction in Pierre Robin sequence.

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Competing interests

None declared.

Ethical approval

Not required.

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