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Mandible traction with wires for the treatment of upper airway obstruction caused by Pierre Robin sequence in Chinese infants: Preliminary findings

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ABSTRACT

Pierre Robin sequence (PRS) is a congenital abnormality that may cause upper airway obstruction requiring surgical intervention. This preliminary study aimed to examine the feasibility and effectiveness of mandible traction with wires for the treatment of upper airway obstruction caused by PRS in Chinese infants. Measures of interest included transcutaneous oxygen saturation before and after surgery, duration of surgery and traction, complications, and CT findings. Seven infants were included in the study (mean birth weight: 2485 g, range: 2405–2570 g); four were born preterm and three were born full term. Mean age at surgery was 13.7 days (range: 2–28 days), mean duration of surgery was 16.6 min (range: 13–25 min) and mean duration of traction was 26.6 days (range: 21–35 days). Mean follow-up was 6.2 months (range: 1–11 months). No infant experienced severe complications. All infants experienced increases in transcutaneous oxygen saturation after surgery. Mean transcutaneous oxygen saturation was 82% before surgery and 98% after surgery. Follow-up morphology of the mandible was excellent. There was no upper airway obstruction, and short-term growth and development were satisfactory. These preliminary findings suggest that mandibular traction with wires may be an effective treatment for upper airway obstruction caused by PRS in Chinese infants.

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1. Introduction

Pierre Robin sequence (PRS) is a congenital condition, characterized by micrognathia and glossoptosis with or without a cleft palate (Scott et al., 2012; van den Elzen et al., 2001), that occurs in approximately 1 in every 8500 to 14,000 live births (Printzlau and Andersen, 2004). The condition, which lacks consensus diagnostic criteria (Breugem and Courtemanche, 2010), may occur alone, or manifest with other syndromes, such as Stickler syndrome, Treacher-Collins syndrome, and velocardiofacial syndrome (Cohen, 1999; Evans et al., 2006; Huang et al., 2007). Clinically, infants with PRS may experience upper airway obstruction and, as a consequence, respiratory distress/dyspnoea, feeding difficulties, and failure to thrive (Mackay, 2011; Scott et al., 2012). The severity of clinical symptoms varies from infant to infant, but may warrant active treatment in some cases (Scott et al., 2012).

Treatment options for PRS may be non-surgical or surgical (Poets and Bacher, 2011). Non-surgical treatment, specifically, placing the infant in a prone or lateral recumbent position, is the first choice option for relieving dyspnoea, but is only effective in 47.6–63.3% of cases (Evans et al., 2006; Schaefer et al., 2004). Moderately invasive treatment options that can be effective, particularly in the short-term (Poets and Bacher, 2011), include establishment of a nasopharyngeal airway, endotracheal intubation, and long-term laryngeal mask use (Abel et al., 2012; Anderson et al., 2007; Evans et al., 2006). For patients who do not respond to non-invasive or minimally invasive treatment, surgical treatment including glossopexy, tracheostomy and mandibular distraction osteogenesis may be considered (Iatrou et al., 2010; Mahrous Mohamed et al., 2011; Poets and Bacher, 2011; Scott et al., 2012). Of these approaches, mandibular distraction osteogenesis is the only treatment that aims to directly address dyspnoea caused by glossoptosis and/or micrognathia. Although these means of treating PRS can be effective, each has limitations/risks, including the need for subsequent surgical procedures (glossopexy), potential trauma and complications (tracheostomy), and potential trauma, scarring nerve injury, and damage to tooth buds (mandibular distraction osteogenesis) (Myer et al., 1998; Poets and Bacher, 2011; Scott et al., 2012). Further,

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there is a lack of consensus as to the optimal surgical treatment for PRS (Mackay, 2011).

Baciliero and colleagues have recently reported a novel surgical approach for treating PRS based on traction of the mandible using wires (Baciliero et al., 2011). Of note, this treatment approach is straightforward and, as reported by the authors, was effective and not associated with any major complications in the treatment of more than 100 patients (Baciliero et al., 2011). To our knowledge, no other reports have confirmed the findings reported by Baciliero et al. (2011). Therefore, we felt wish to share our experience and the findings from a preliminary investigation examining the feasibility and effectiveness of mandible traction with wires (slightly modified from the approach described by Baciliero and colleagues) for the treatment of upper airway obstruction caused by PRS in a small cohort of Chinese infants.

2. Materials and methods

2.1. Patients

This study involved infants with PRS treated at the Children's Hospital of Fudan University between June 2011 and June 2012. The diagnosis of PRS was based on morphological abnormality, including micrognathia and glossoptosis, with or without a cleft palate, in addition to upper airway obstruction and difficulties in respiration and feeding. Glossoptosis and/or pharyngeal occlusion were confirmed by computed tomography of the oropharynx (sagittal view). Infants were included in the study if they received treatment via mandible traction with wires (details ensure). Infants with mild symptoms were excluded from the study because surgical intervention was not indicated. All infants underwent chest X-ray after admission to rule out other potential causes of dyspnoea, such as pulmonary infection, atelectasis, and pneumothorax. None of the infants exhibited any sign of airway narrowing on three-dimensional airway reconstruction after facial and chest computed tomography scans. None of the infants had received any prior surgical intervention for the treatment of PRS.

This study was approved by the Institutional Review Board of Children's Hospital of Fudan University. All parents of infants provided written informed consent before surgical procedures were commenced.

2.2. Surgery

The surgical procedure described by Baciliero et al. (2011), which is based on a technique first reported by Stellmach and Schettler (1968), was used. Surgery was performed if the mean transcutaneous oxygen saturation was less than 90% after a 24-h monitoring period in the lateral/prone position, or if the transcutaneous oxygen saturation decreased continuously due to dyspnoea during the monitoring period and manual intervention/rescue was required.

Local anaesthesia was applied to the chin. A trocar was then inserted from a point 0.5 cm distal to the middle of the chin at the inferior margin of one side of the mandible. The trocar was advanced under the periosteum close to the medial side of the mandible and out through the floor of the mouth. The needle core was pulled out, a wire was placed through the trocar, and the trocar was removed. This strategy helps to prevent injuring muscles in the base of the tongue. Another trocar was then inserted under the periosteum anterior to the mandible and the gum (against the outlet of the wire from the floor of the mouth) and advanced through the original inlet in the chin. The wire from the floor of the mouth was placed in the trocar, and the trocar was pulled out to complete wire placement on one side. The same approach was used for wire placement on the other side. Bilateral wire insertion points were kept as symmetrical as possible (Fig. 1). Wires on the two sides were connected by a small wooden block to balance the traction force.

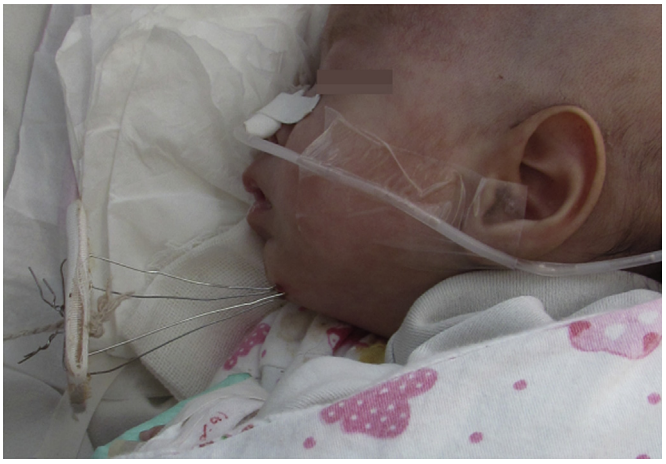

Fig. 1. Photo illustrating traction wires connected to a wooden block to balance the traction force on both sides of the mandible.

After surgery, the patient was transferred to the neonatal intensive care unit and traction (maintained at 100 g) was initiated with the patient in a lateral recumbent position (Fig. 2) as previously described (Baciliero et al., 2011). Transcutaneous oxygen saturation was monitored. The direction of the patient's lateral recumbent position was changed and oral care was provided every 2 h. Postoperative intravenous antibiotics were given for two days. No anaesthesia was given during traction, which the patients underwent without obvious distress. Infants were fed via a nasogastric tube during traction. During this time, constant oral care was provided to prevent the accumulation of oral secretions.

Traction was permanently stopped when the transcutaneous oxygen saturation remained above 95% for at least three days without traction, and when oropharyngeal computed tomography revealed an unobstructed pharynx.

2.3. Measures

Measures of interest in this study included transcutaneous oxygen saturation before and after surgery, duration of surgery, traction, and hospital stay, complications, body weight at discharge, and CT findings. Subjective assessments of mandibular morphology, growth and development, and respiratory function were made during follow-up after discharge.

3. Results

Eleven infants with PRS were treated in our hospital between June 2011 and June 2012. Of these infants, four had mild symptoms and did not require surgical intervention. Hence, seven infants who received mandible traction with wires were included in the study.

The infants' demographic characteristics and surgical details are summarized in Table 1. Four of the infants were boys and three were girls. The mean birth weight was 2485 g (range: 2405–2570 g). None of the PRS infants had an associated syndrome. Four of the infants were born preterm and three were born full term. The mean age at the time of surgery was 13.7 days (range: 2–28 days). The mean duration of surgery was 16.6 min (range: 13–25 min) and the mean duration of traction was 26.6 days (range: 21–35 days). The mean weight at discharge was 3480 g (range: 3050–3650 g)

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