



Tongue-lip adhesion

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The management of Pierre Robin sequence is challenging and requires a multidisciplinary approach. A thorough workup must be done to exclude sites of airway obstruction other than the base of the tongue and to exclude central sleep apnea. Airway security is critical and can be managed nonoperatively in most children. When surgical management is necessary, the tongue-lip adhesion is the first step in many institutional algorithms. Multiple modifications have been described, but the basic premise is an anterior advancement of the tongue base and suture fixation of the tongue to the lip. This increases the diameter of the airway and results in safe and successful management in the overwhelming majority of patients. There are few sequelae with the most common being dehiscence. This can be prevented by intermuscular sutures, a circummandibular stitch, and release of the genioglossus muscle. Once mandibular “catch-up” growth has been achieved, the tongue-lip adhesion may be reversed.

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Introduction

The clinical triad of micrognathia, glossoptosis, and airway obstruction is referred to as Pierre Robin sequence. Although our understanding of the etiology is still incomplete, it is generally accepted that Pierre Robin represents a sequence and not a distinct syndrome as the clinical associations are the result of a prior developmental anomaly. In Pierre Robin sequence, a symmetric and hypoplastic mandible (micrognathia) forces the tongue to be posteriorly displaced which creates glossoptosis resulting in airway obstruction (Figure 1).

Pierre Robin sequence may occur in isolation or in conjunction with other syndromes such as Stickler, 22q11 deletion, fetal alcohol, Nager syndrome, or Treacher Collins syndrome. The incidence of isolated Pierre Robin sequence is reported to range from 17%–63%.^{1,2} U-shaped clefting of the secondary palate occurs in approximately 90% of patients with Pierre Robin sequence owing to malposition

of the tongue which creates a mechanical obstruction during fusion of the palatal shelves. Although there is debate in the current literature whether cleft palate is a universally defining feature, airway obstruction is always present to a degree and is the primary determinant of morbidity and mortality.

Early recognition and management of Pierre Robin sequence is essential to preventing devastating complications associated with airway obstruction including feeding difficulty, aspiration, failure to thrive, chronic hypoxemia, elevated pulmonary vascular resistance, cor pulmonale, and death.^{3,4} In Robin’s era, the mortality rate was as high as 65% by 18 months of age owing to cachexia and pulmonary infection.⁴ Over the past 2 decades, as multidisciplinary management protocols have been implemented, the mortality rate has significantly decreased to 4%–26%.² The mortality rate in isolated Pierre Robin sequence has been reported to be 5.9% with the rate rising to 22.8% in syndromic patients.⁵

When treating a child with Pierre Robin sequence, the first priority is properly addressing the airway. Prone positioning employs gravity to relieve the posterior displacement of the tongue which can obstruct the oropharynx. This simple nonsurgical modality has been

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Figure 1 Preoperative view demonstrating glossoptosis and cleft palate. (Color version of figure is available online.)

reported to successfully treat airway obstruction in 50%-70% of patients with Pierre Robin sequence.^{3,6} Although prone positioning may relieve the obstruction in most patients, silent obstruction may still occur, and ongoing surveillance is mandatory.

If an infant fails prone positioning, a nasopharyngeal airway, with or without a Kirschner wire for stabilization, is another nonsurgical option.^{7,8} In a recent study by Meyer, 48% of children who required an airway intervention were successfully treated with a nasopharyngeal airway. In 1 series, those treated with nasopharyngeal airways required continuous pulse oximetry and a prolonged hospital stay (mean = 60 days, range: 25-162 days).⁷ Others have reported a 75% success rate with nonsurgical modalities.⁹

Tongue-lip adhesion, or glossopexy, is a well-established part of the surgical algorithm for airway obstruction in Pierre Robin sequence. The first rudimentary description of the tongue-lip adhesion is credited to Shukowsky¹⁰ in 1911, several years before Pierre Robin's landmark article in 1923 which linked the clinical triad that bears his name.¹¹ Douglas¹² formally described the tongue-lip adhesion by suturing the anterior portion of the tongue in a forward position in 1946. Since that time, there have been modifications to the technique and debate regarding the role of tongue-lip adhesion in the management of Pierre Robin sequence. Modifications have included intermuscular sutures, an anchoring stitch around the anterior mandible, a retention button over the base of the tongue, and division of the genioglossus muscle for anterior mobilization of the tongue. Independent of the technique, tongue-lip adhesion has a reported success rate ranging from 83%-100%.^{3,13}

Mandibular distraction osteogenesis has also been advocated for the treatment of Pierre Robin sequence. In this technique, bilateral osteotomies are made in the mandible and an external or internal distraction device gradually lengthens the mandible, which increases the pharyngeal airway size.¹⁴⁻¹⁶ Although the procedure is safe, the distraction progresses at a rate of

0.5-2 mm per day. As such, the airway must be secured during the process with an endotracheal tube, tracheostomy, or nasopharyngeal airway. Potential complications include damage to the marginal mandibular and inferior alveolar nerves, injury to premolar tooth buds, infection, and dislodgement of the distractor.¹⁵ The decision to use distraction osteogenesis vs tongue-lip adhesion appears to be based on surgical training and philosophy.

Regardless of the philosophy, there is agreement that tracheostomy is considered a last resort. Tracheostomy may be indicated in patients who have failed to respond to other measures or when there are multiple levels to the airway obstruction.

Indications

Surgical treatment of Pierre Robin sequence has the potential for severe morbidity and should only be explored when conservative methods have been exhausted. There is significant heterogeneity between the anatomy, clinical presentation, and severity of patients with Pierre Robin sequence, even when comparing isolated or syndromic patients. As such, it is imperative that a thorough workup of airway obstruction is undertaken to isolate the etiology.

A complete flexible nasopharyngoscopy and bronchoscopy evaluation is mandatory. There may be multiple levels of obstruction or other pathologic anomalies such as tracheomalacia, acute angulation of the basicranium, subglottic anomalies, or hypotonia. Anterior mobilization and adhesion of the tongue will not correct these other underlying pathologies and is not indicated in these mixed clinical presentations.

Polysomnography is an important study used to differentiate between central and obstructive sleep apnea. A polysomnogram consists of a 24-hour recording of a nasal thermistor, oral thermistor, end-tidal carbon dioxide, thoracic and abdominal strain gauges to monitor mechanical respiratory effort, electromyography of the phrenic nerve, pulse oximetry, electrocardiography, and tachography. Central sleep apnea is diagnosed when no muscle effort is associated with apnea, and conversely obstructive sleep apnea is diagnosed when there is muscle effort in the absence of detected airflow.

Technique

Tongue-lip adhesion is performed under general anesthesia preferably via nasotracheal intubation. In our institution, patients are given a preoperative weight-based dose of intravenous clindamycin for bacterial prophylaxis as well as dexamethasone to minimize postoperative edema. Deep venous thromboembolism prophylaxis is not indicated.

Once the patient is properly anesthetized, local anesthesia (1:1 mix of 0.25% bupivacaine and 0.5% lidocaine with 1:200,000 epinephrine) is infiltrated at the base of the tongue, submentally and along the lingual and buccal aspect of the mandible.

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