



The tongue-lip adhesion

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The tongue-lip adhesion (TLA) has long been a part of the management algorithm for glossoptosis associated with the Pierre Robin sequence (PRS). Advancement of the base of the tongue has been shown to relieve airway obstruction in some infants. TLA has few complications or long-term sequelae. The procedure has also been shown to ameliorate the feeding difficulties associated with PRS and therefore, reduces hospital stay, simplifies nursing care, and makes home care less demanding.
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The tongue-lip adhesion (TLA) has long been a part of the management algorithm for glossoptosis associated with the Pierre Robin sequence (PRS). First described in its rudimentary form in 1911 by Shukowsky, the procedure has since undergone appreciable changes in both technique and philosophy of implementation.¹ The principle behind the procedure remains the same. It is essentially an advancement of the tongue, specifically of the base of the tongue, to relieve its collapse and impingement on the posterior oropharynx (Figure 1). While some may argue about its utility, the glossopexy, as it is also referred to in the literature, carries with it fewer complications and long-term sequelae than either tracheostomy or mandibular distraction. Several authors have reported success with the procedure despite variations in technique.¹⁻³ A number of case series have been published highlighting the applicability of TLA.¹⁻⁴ It has successfully been shown to relieve airway obstruction in infants with PRS that is refractory to positioning alone.³ The procedure has also been shown to ameliorate the feeding difficulties associated with PRS and, therefore, reduces hospital stay, simplifies nursing care, and makes home care less demanding.⁵

The procedure involves the creation of a wound on the ventral surface of the tongue that is to adhere to a wound created on the lower lip. This adhesion can be anchored or

supported in various ways including the use of sutures connecting the muscles within the tongue and lip,^{2,3} the use of an anchoring stitch around the anterior mandible, and the use of a retention button over the posterior dorsum of the tongue.¹ Stripping of the origins of the genioglossus has also been advocated by some, to aid in mobilization of the tongue anteriorly.² Regardless of the technique employed, it is essential to determine the underlying cause of airway obstruction in PRS. A thorough upper and lower airway examination is mandated as a significant percentage of infants with PRS have a syndrome and the causes of apnea in these infants may be heterogeneous.⁶ There may be multiple levels or mechanisms of airway obstruction in which case anterior mobilization of the tongue alone would be rendered ineffective.

Indications

Before surgical intervention is considered, nonsurgical management should be attempted. Management algorithms begin by evaluating the effectiveness of prone positioning.^{1-4,6-8} Nasopharyngeal airway (NPA) placement can also be attempted should positioning fail.^{4,6-8} Sher⁶ has reported that at least 50% patients treated with NPAs did not require definitive surgical intervention to correct airway obstruction. Wagener et al⁸ and Anderson et al⁹ had no failures with their nonsurgical management protocol using an NPA in the Birmingham unit.

It is critical that glossoptosis be determined as the definitive etiology for airway obstruction in a PRS infant before a TLA is performed. Sher⁶ described 4 different mecha-

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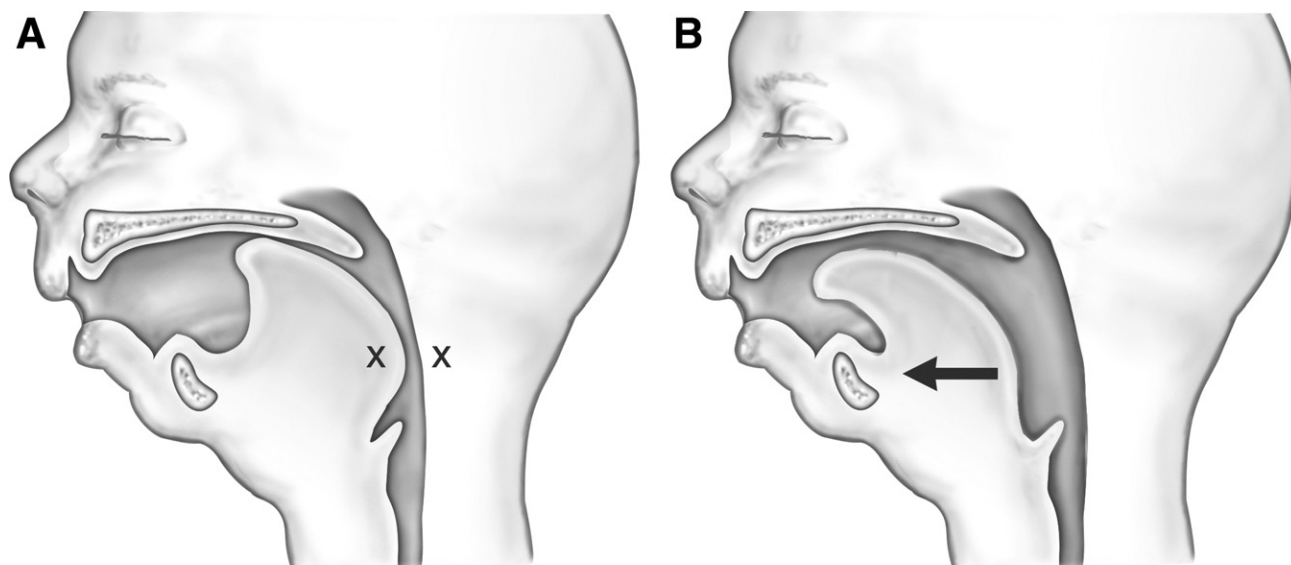


Figure 1 (A) Glossoptosis associated with Pierre Robin sequence results in upper airway obstruction. (B) The posterior tongue is advanced and fixed forward with some form of anchorage. This effectively relieves the glossoptosis and the resultant airway obstruction.

nisms of airway obstruction in patients with craniofacial anomalies, including PRS (Figure 2). The TLA has been shown to be effective in addressing Type 1 obstructions defined as posterior movement of the dorsum of the tongue to the posterior pharyngeal wall so that most of the obstruction is anteroposterior.^{1,6} Flexible fiberoptic nasopharyngoscopy can be used to elucidate this as the underlying etiology of airway obstruction and this determination can therefore guide surgical management.^{1,6}

Technique

The TLA is performed under general anesthesia. Local anesthesia is infiltrated (0.5% lidocaine with 1:200,000 epinephrine). A transverse incision is made on the ventral surface of the tongue just above the floor of the mouth. Short perpendicular releases are made at either end of the transverse incision and are directed toward the floor of the mouth. Care must be taken to visualize the openings of the submandibular and

Type 1: Posterior movement of the dorsum of the tongue to the posterior pharyngeal wall so majority of obstruction is anteroposterior.

Type 2: Posterior movement of tongue causing compression of the soft palate or cleft palatal tags posteriorly against the posterior pharyngeal wall.

Type 3: The lateral pharyngeal walls move medially causing them to oppose one another.

Type 4: The pharynx constricts in a circular or sphincteric manner with movements occurring in all directions.

Figure 2 Mechanisms of upper airway obstruction in patients with craniofacial anomalies.

sublingual gland ducts. It is through this incision that the genioglossus can be stripped, though not all authors advocate this approach. A transverse incision is then made in the mucosa of the lower lip at the base of the labial vestibule and the incision is released perpendicularly at either end toward the lip (Figure 3). Both flaps should be deep to muscle. The flaps are then brought together such that the transverse incision on the ventral surface of the tongue is brought forward and sutured to the transverse incision at the base of the labial vestibule of the labial flap. This creates a continuous raw surface for adhesion (Figure 4). Once the inner flap of the adhesion is sutured, deep sutures are used to bring together the intrinsic muscles of the tongue and the orbicularis oris. It is universally agreed that this is a necessary step in the procedure to avoid wound dehiscence.¹⁻⁴ As mentioned earlier, several variations on the technique have been reported. Kirschner et al³ advocate the use of a retention button on the dorsum of the tongue that is anchored below the anterior chin (Figure 5), and this variation has been successful, reporting few complications. Argamaso¹ has reported similar results, advocating the use of a suture looped around the anterior mandible and tied to the intrinsic muscles of the tongue. It is advisable that after the procedure is complete, endoscopic evaluation be undertaken to observe relief of glossoptosis while the infant is supine.^{1,3} Most authors recommend a nasopharyngeal tube during the immediate postoperative period, to be left in place for 2-3 days.¹⁻⁴ The patient may then remain in the semirecumbent position while awake. Adequate gas exchange must be verified with continuous pulse oximetry or formal sleep study as mandated clinically.⁶ Nasogastric feeding should continue for approximately 1 week to allow healing and to avoid compromising the wound by vigorous suckling. Once oral feeding begins, and caloric intake is appropriate for daily requirements, the patient may be discharged. Frequent

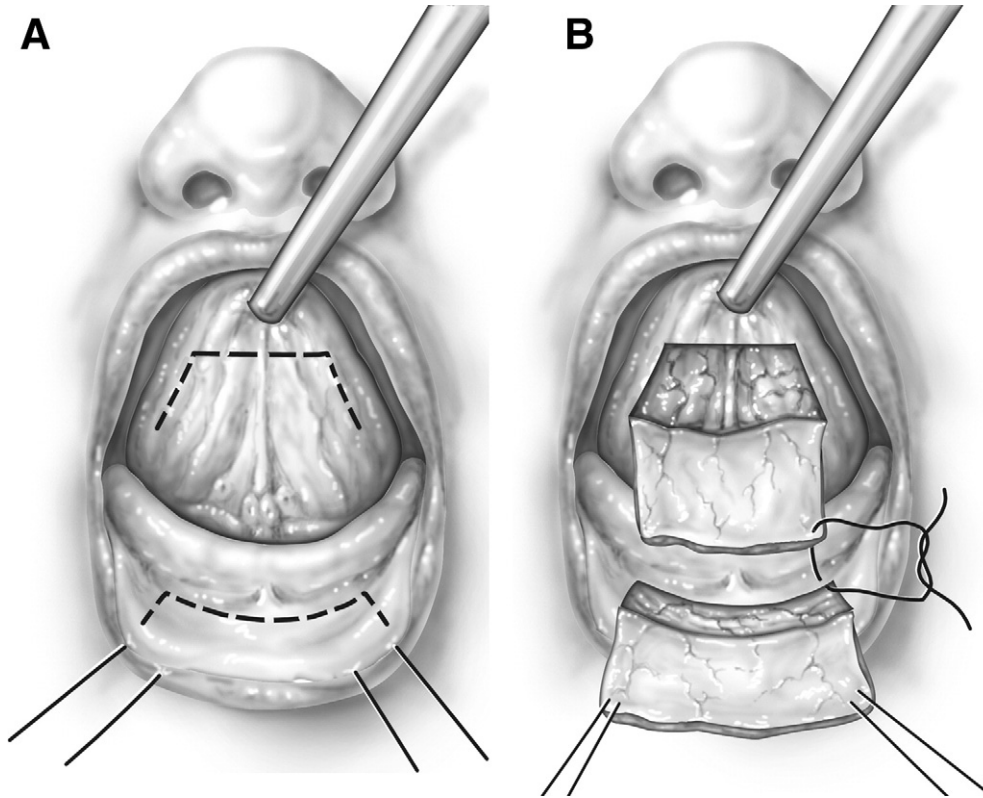


Figure 3 (A) Transverse incisions are made on the ventral surface of tongue and lower lip. These are carried deep to muscle. (B) The leading edge of the mucosal tongue flap is carried to the proximal base of the lip incision where they are sutured to begin the mucosal approximation.

follow-ups to monitor growth and development and nasopharyngoscopic examinations to determine airway patency are warranted.⁶

Timing of tongue release

There is no general consensus as to when the tongue should be released. Most authors agree that it should be done at or before repair of the palate.^{1-4,10} Proponents of earlier takedown argue that it is essential to determine if the infant can maintain a patent airway after palatal repair.³ Takedown of the adhesion earlier also facilitates proper hygiene of lower central incisors.³ Proponents of the simultaneous takedown and palate repair appear to base their decisions solely on avoiding multiple exposures to anesthesia and hospitalization.¹

Complications

Largely because of modifications in incision design by Routledge in 1960, few complications have been reported in the literature, all of which are fairly uncommon.² Dehiscence is the most commonly noted complication occurring in 4.2%-17.2% patients.¹⁻⁴ Abnormal eruption of deciduous incisors has also been reported.¹ Though not a complication, some authors have reported a failure rate as high as 20% in the procedure. This is likely attributable to inaccurate diagnosis of the cause of airway obstruction. Contrary to popular belief, given the young age of adhesion takedown, speech development is generally not adversely affected by the procedure.^{1,2,10} Scarring that results from the TLA is usually minor and of no esthetic or functional consequence.

Figure 4 Profile view indicating where the edges of the flap are brought together to facilitate the raw surface to raw surface interfaced adhesion.

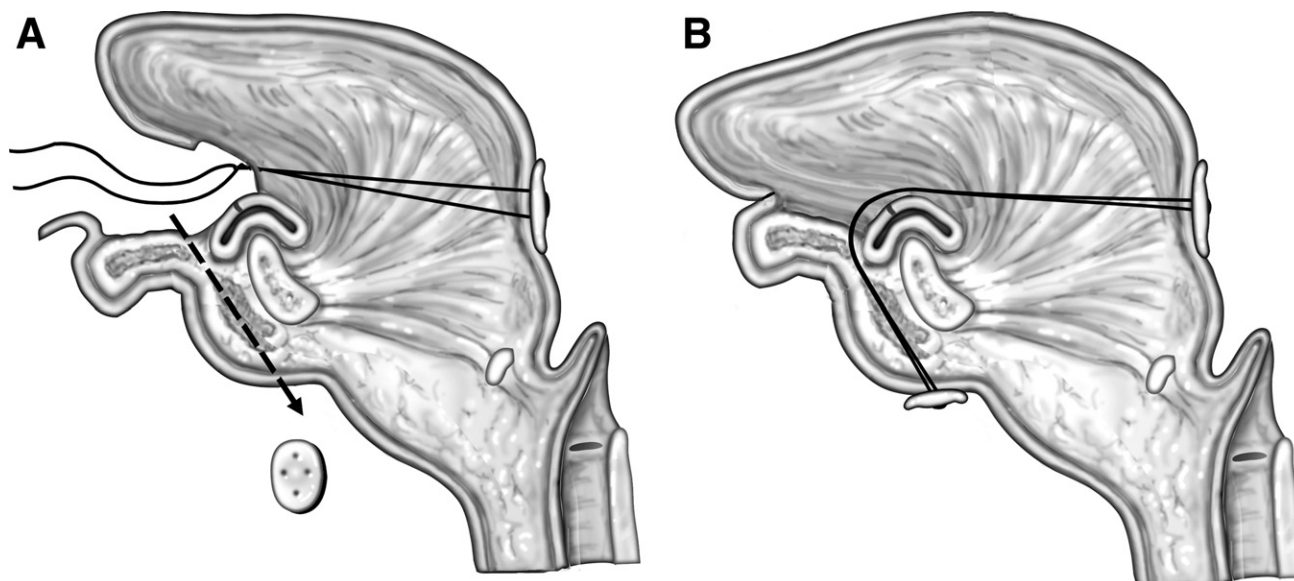


Figure 5 (A) A retentive button is tacked to the posterior base of tongue. (B) The suture is then brought through raw wound and then down and anterior to the mandibular alveolus where it is anchored to another retentive button under the chin.

Discussion

Although limited in its applicability, the TLA has a definitive role in the management of upper airway obstruction in infants with PRS. The evolution of the procedure over the last 60 years is a function of ingenuity in minimizing the complications of earlier techniques and, more importantly, in addressing the etiology for which the procedure is effective at circumventing—glossoptosis. It should be emphasized that a thorough history and physical examination, including genetic testing, is essential to rule out a syndromic diagnosis in children with PRS, which is found in isolation in only 20% of cases.⁶ Syndromic infants have poorer outcomes and are more likely to have persistent apnea after TLA despite nasopharyngoscopic confirmation of glossoptosis.^{1,3} This is likely a reflection of the heterogeneous nature of upper airway obstruction in PRS. Hypotonia secondary to neurologic impairment will work synergistically to impair posterior pharyngeal obstruction. That said, the TLA is a relatively easy, reversible procedure with fewer long-term complications than either mandibular distraction or tracheostomy.

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