

# Tongue–lip adhesion in Pierre-Robin sequence: Role redefined

### ABSTRACT

The triad of retrognathia, glossoptosis, and airway obstruction characterizes the Robin sequence along with the detrimental effects of mandibular hypoplasia on feeding, swallowing, and growth, which are very well described. Most of the babies are managed successfully on nonsurgical measures, but selected patients require surgical intervention in the neonatal period for survival. Conventionally, tracheostomy was done, which still remains a first-line surgical procedure for some surgeons. However, presently, most of the craniofacial centers have switched over to mandibular distraction procedures at an early stage and only sometimes tongue–lip adhesion (TLA). The literature is unclear as to which surgical procedure for securing the airway is more effective for these patients, and hence, the choice of procedure depends on the resources and surgical expertise. This article tells the tale of a neonate who survived by just placing a simple U-stitch between the tongue and lip, retracting the tongue outside, which is the basic concept of all TLA procedures. It also reemphasizes the importance of TLA in Robin patients to improve the airway obstruction and helps buy the time in which the mandible and associated structures grow.

**Keywords:** Airway, micrognathia, Pierre-Robin sequence, tongue–lip adhesion

### INTRODUCTION

Pierre-Robin sequence (PRS) has been classically defined as micrognathia, glossoptosis, and upper airway obstruction, with or without a cleft palate. This sequence frequently contributes to feeding problems which may delay oral feeding. The condition is commonly associated with other craniofacial anomalies, further complicating the airway management.<sup>[1]</sup>

Tongue–lip adhesion (TLA) has been used in the treatment of airway obstruction in PRS ever since popularized by Douglas in 1946.<sup>[2]</sup> Since its inception, it has undergone several modifications by Routledge,<sup>[3]</sup> Randall,<sup>[4]</sup> and Argamaso<sup>[5]</sup> in hope of reducing the surgical complications, namely dehiscence and possible failure. Some other modifications such as posterior placement of the tongue incision just above the floor of the mouth along with stripping of the origins of the genioglossus muscle and use of tongue suspension in conjunction with a standard transverse adhesion of the lip have also been described.<sup>[6,7]</sup> The indications include patients

with type 1 or 2 airway obstructions, who experienced desaturation when nursed in the supine position or during feeding but remained very much stable when the prone position is maintained. Through this article, we want to convey that in spite of all the modifications, even a simple U-stitch can serve the purpose of TLA.

### CASE REPORT


A full-term male neonate born with PRS was referred from elsewhere neonatal care unit to our center with

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severe respiratory distress and bluish discoloration of the peripheries. The neonate was admitted in the neonatal intensive care unit (NICU) under plastic surgery and initially nursed in the prone position on 100% oxygen through mask and Ryle's tube feeding. Continuous monitoring was done for maintaining the saturation at 98%. This status continued for 15 days. After 2 weeks, a trial was made to keep the neonate in the lateral position which he could not tolerate and again nursed in the prone position. Another 2 weeks passed by, but there was no improvement during the change of posture. Hence, a team, including a plastic surgeon, ear, nose, and throat specialist, pediatrician, and anesthesiologist decided to assess the airway under general anesthesia and look for any mechanical causes of obstruction. It was also decided to do a procedure for preventing the tongue fall, if possible.

Under general anesthesia, a flexible laryngoscope was used to assess the airway, which was found to be normal. Since other causes of mechanical obstruction were excluded, a decision was made to perform TLA procedure. The mucosa of the tongue was infiltrated with a minimal amount of lidocaine 0.25% with epinephrine 1:400,000. Since the baby was very small and his safety was of prime concern, we decided not to do the extensive procedure of TLA requiring the elevation of flaps and suturing them together. Instead, using a heavy-needle holder, no. 1 Mersilk on a large needle was passed through the lower lip under the chin. The suture was passed over the mandible and then through the anterior aspect of the tongue and brought back through the tongue as a loop. This U-stitch was again passed through the lower lip and tied under the chin over bolsters to avoid pressure necrosis [Figure 1]. At the completion of the procedure, we ensured that the tongue protruded through the space between the lips. The endotracheal tube was left in place for 24–48 h. Vigorous

monitoring in NICU was done, and the tube was removed once the infant was able to breathe normally.

Daily dressing of the bolster stitch was done, and care was taken for infection and cut-through of the stitch. The child was nursed in the supine position without oxygen supplementation. There was a problem of excessive salivation which was managed by frequent change of dressing. This stitch was kept in place for 1 month and gradually loosened while monitoring the oxygen saturation and finally removed. At that time, the child was 2 months old. Few days later, Ryle's tube was also removed, and the patient was discharged in a stable condition. The mother was taught to observe the child for any signs of respiratory distress or blueness. Follow-up was done at weekly interval for 4 weeks, every 2 weeks for 3 months, and then monthly. Scarring on the lip has been insignificant, and there was no visible scar on the tongue [Figure 2].

## DISCUSSION

Pierre-Robin anomalad is a challenging deformity. Robin sequence is a heterogeneous condition caused by isolated mandibular hypoplasia, or by a multitude of conditions that can produce a similar phenotype. More than 50% of affected patients have associated syndrome, genetic, or medical anomaly.<sup>[1]</sup> Most babies born with it are successfully managed by nonsurgical techniques. With the development of new-born intensive care units, the TLA procedure is needed only for an occasional infant whose respiratory problems are refractory to techniques of positioning, nasogastric intubation, nasopharyngeal airway, and temporary endotracheal intubation.

TLA is the most conservative step available for the management of those few infants with Pierre-Robin anomalad



Figure 1: Intraoperative photograph showing bolster tie over for tongue–lip adhesion



Figure 2: Three months postoperative photograph of the child with Pierre-Robin Sequence

who have persistent respiratory obstruction after an adequate trial of nonsurgical management. Tracheostomy is reserved for those rare patients who have complications or failure of TLAs, which cannot be otherwise managed. Infants undergoing TLA may have dehiscence, infection, submaxillary duct obstruction, lip scarring, postoperative obstructive sleep apnea, severe dysphagia requiring prolonged gastrostomy tube usage, and growth retardation with a high rate of secondary procedures performed. The complication rate has been reported from 17% to 43%.<sup>[8]</sup>

To conclude, the placement of a simple U-stitch between the tongue and lower lip in PRS patients not responding to conservative measures can also act as a good and durable alternative to standard TLA procedures.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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#### Conflicts of interest

There are no conflicts of interest.

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