

피에르 로빈 연속증의 치료로써 치조 보호 장치를 이용한 혀-하순 유착술

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Tongue-Lip Adhesion Using an Alveolar Protector Appliance for Management of Pierre Robin Sequence

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Purpose: Pierre Robin sequence is a congenital malformation in which micrognathia causes glossoptosis and airway obstruction. If conservative treatment fails, surgical procedures such as tongue-lip adhesion can be performed. However, this procedure remains a subject of debate, with favorable results being countered by reports of complications. To overcome the above limitations, we revised the traditional method of tongue-lip adhesion using an alveolar protector.

Methods: Between 1992 and 2011, a total of eight patients were identified with Pierre Robin sequence and were treated with tongue-lip adhesion. Two of these eight tongue-lip adhesion procedures were performed with an alveolar protector. The operative technique for tongue-lip adhesion was similar to that described in other published reports. The alveolar protector was inserted between the ventral surface of the tip of the tongue and the lower labial sulcus.

Results: Tongue-lip adhesion failed in two patients because of wound dehiscence. The primary surgical success rate was 66.7%. In the two tongue-lip adhesion procedures performed with the alveolar protector, we observed no postoperative complications.

Conclusion: Resistance to traction of the tongue can be encountered with nonunionized symphysis menti, causing loosening of the traction suture through the symphysis menti. This can lead to backward positioning of tongue, resulting in dehiscence of tongue lip adhesion. The

alveolar protector is a good adjunct to tongue-lip adhesion because this method avoids postoperative loosening of the traction suture and wound dehiscence. It is a simple and effective auxiliary method that yields functional improvement.

Key Words: Pierre Robin sequence, Tongue-lip adhesion, Alveolar protector, Treatment

I. INTRODUCTION

Pierre Robin sequence is classically defined as the triad of micrognathia, glossoptosis, and upper airway obstruction, with most infants also having a cleft palate.¹ Infants frequently have feeding and respiratory difficulties caused by posterior displacement of the tongue due to mandibular hypoplasia.

There are well-known treatments for airway obstruction, including position change, tongue-lip adhesion, distraction osteogenesis of the mandible, and tracheostomy. Initially, the condition can be managed in infants by placing them in the prone position and using gastric lavage feeding tubes. If the infant does not improve with change in position, an oral airway or nasopharyngeal intubation may relieve distress. If conservative treatment fails, surgical procedures such as tongue-lip adhesion can be performed. Although tongue-lip adhesion is a simple and effective surgical procedure, it has been reported to be associated with postoperative complications, including dehiscence and injury to the adjacent structures. Wound dehiscence occurred in 17% to 42% of patients.² Due to this frequent occurrence of dehiscence and the frequent necessity of revisions, tongue-lip adhesion has gradually been abandoned.¹

Tongue-lip adhesion can be effective in relieving tongue-based airway obstructions in the neonatal period. However, this procedure remains a subject of debate, with favorable results being countered by reports of postoperative complications such as wound dehiscence and loosening of the traction suture. To overcome the aforementioned limitations, we revised the traditional

Received March 16, 2011

Revised April 25, 2011

Accepted April 26, 2011

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method of tongue-lip adhesion using an alveolar protector.

II. IDEAS AND INNOVATIONS

From October 1992 to January 2011, a total of eight patients were diagnosed with Pierre Robin sequence and treated with tongue-lip adhesion at our institute. Two of these eight tongue-lip adhesion procedures were performed with an alveolar protector (Table I).

Tongue-lip adhesion without an alveolar protector failed in two patients because of wound disruption within a week. The primary surgical success rate was 66.7%, which was consistent with other published reports.²

All patients were admitted to the pediatric neonatal intensive care unit with upper airway obstruction, difficult feeding, and poor body weight gain. Our initial treatment for patients with Pierre Robin sequence was conservative treatment, including position change, instrumental airway maintenance, oxygen supply, and nutritional support. When conservative treatment failed to treat the airway obstruction, surgical treatment was performed. Tongue-lip adhesion was performed at a mean age of 59 days old, ranging from 13 to 126 days.

This method is a simple modification of the conventional tongue-lip adhesion introduced by Douglas.³ The procedure was performed under general anesthesia. To create the alveolar protector, we mixed a piece of viscous vinyl polysiloxane material (3M Express STD Putty; 3M Dental Products). It was inserted into the patient's mouth and the material was pressed over the lower labial sulcus. When the material was molded into the desired form, it was removed from the mouth and hardened extraorally. The protector was designed surrounding the alveolus, with a length of 15~20 mm, a width of 5~8 mm,

and a depth of 1~2 mm (Fig. 1).

The operative technique used for the tongue-lip adhesion was similar to that described in other published reports. Two mucosal flaps of the same size (20 × 5 mm) were designed on the ventral side of the tongue and on the inner side of the lower lip (Fig. 2). After infiltration

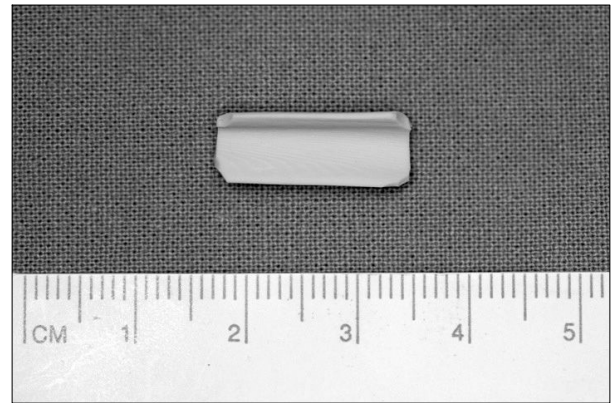


Fig. 1. The alveolar side view of protector.

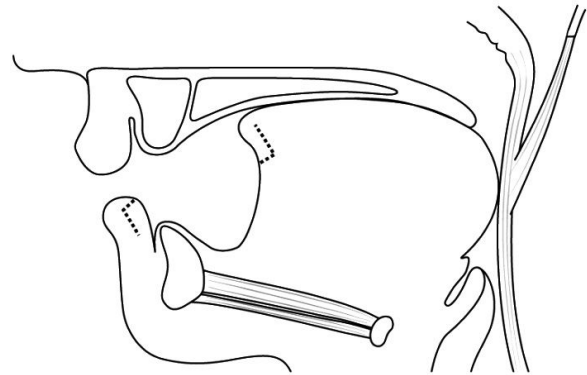


Fig. 2. Two mucosal flaps of the same size were designed on the ventral side of the tongue and on the inner side of the lower lip.

Table I. 8 Pierre Robin Infants with Tongue-lip Adhesion: A Summary of Characteristics and Outcomes

Patient	Date of birth	Sex	Operation	Age at operation (Days)	Dehiscence	Detachment	Outcome
1	10 /6/1992	M	TLA	49	-	POD#302	No apnea
2	9/1/1994	M	TLA	126	-	POD#355	No apnea
3	3/3/2006	F	TLA	18	POD#5	-	No apnea
4	3/21/2007	F	TLA	13	-	-	Expired
5	6/19/2007	F	TLA	126	-	POD#485	No apnea
6	11/4/2009	M	TLA	54	POD#6	-	No apnea
7	1/11/2010	M	TLA-AP	53	-	POD#528	No apnea
8	1/10/2011	M	TLA-AP	33	-	Maintain	No apnea

TLA, Tongue-lip adhesion; TLA-AP, Tongue-lip adhesion with alveolar protector.

with lidocaine 1% with epinephrine 1 : 100,000, incisions were made below the tip of the tongue and in the wet mucosa of the lower lip. The horizontal incision in the lower labial sulcus should be placed a few millimeters superior to the gingival sulcus. The corresponding horizontal incision in the tongue should be placed on the ventral surface of the tongue and measure 20 mm in length, to provide adequate surface area for adhesion. An inferior-based mucosal flap from the lower lip and a superior-based mucosal flap from the ventral surface of tongue were elevated, exposing the underlying muscle of the lower lip and tongue. The ventral surface of the tongue was pulled anteriorly to contact the lower lip mucosa. Slight tension was exercised with this action to move the tongue base forward and release the pharyngeal airway (Fig. 3). The alveolar protector was inserted between the ventral surface of the tongue tip and the lower labial sulcus (Fig. 4). Retention of the appliance

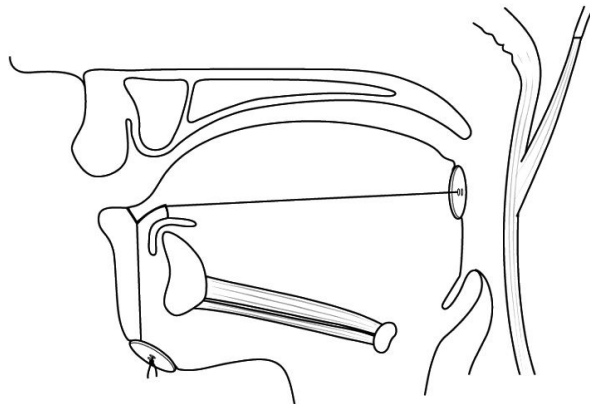


Fig. 3. The ventral surface of the tongue was pulled anteriorly to contact the lower lip mucosa. Slight tension was exercised with this action to move the tongue base forward and release the pharyngeal airway.

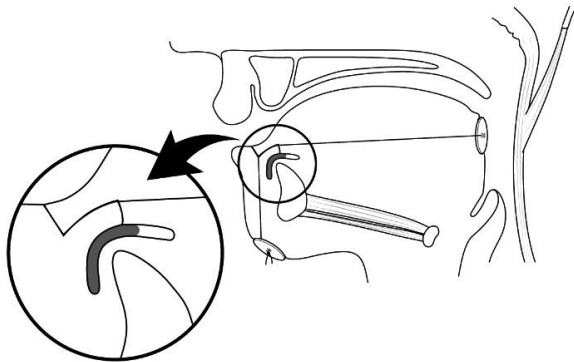


Fig. 4. The alveolar protector is a prosthesis that is designed to restore separation between the ventral surface of the tongue tip and the lower labial sulcus.

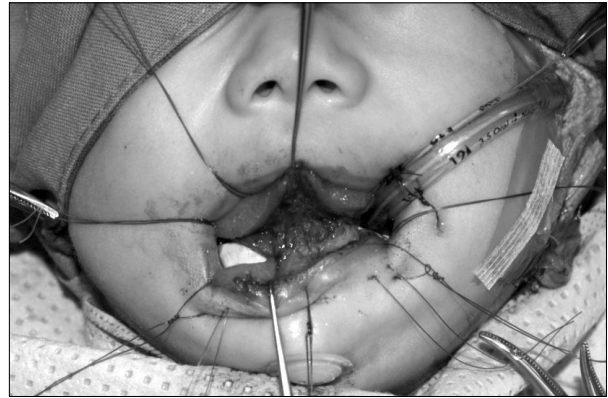


Fig. 5. Intraoperative view showing tongue-lip adhesion with the alveolar protector appliance.

was performed using 3-0 Nylon sutures (Fig. 5). The lower lip mucosal flap was flipped backwards across the gum and sutured to the lower edge of the tongue wound. Muscle to muscle approximation between the muscularis propria of the tongue and orbicularis oris muscle was achieved using 5-0 Vicryl sutures. A horizontal mattress traction suture of Nylon 2-0 was passed from the tongue base through the tongue musculature and exited via the ventral surface of the tongue tip. The suture was then brought down towards the lower labial sulcus, anterior to the mandibular symphysis, and brought out through the chin skin. This suture connected the button on the posterior surface of the tongue to the other button beneath the chin. The Nylon traction suture was tied to the point at which the base of the tongue begins to move forward. Tightening was maintained in order to prevent breathing difficulty from retracting the tongue backwards. Finally, the tongue mucosal flap was then flipped forward and sutured to the superior edge of the lower lip wound, covering the tongue-lip muscle sutures.

The alveolar protector was kept in for 3 weeks and used to protect the wound during healing. After confirming upper airway patency, the tongue-lip traction suture and the buttons were removed.

III. DISCUSSION

The Pierre Robin sequence is a congenital anomaly with characteristics of micrognathia, glossoptosis, and upper airway obstruction.⁴ Clinical symptoms during infancy are difficulty breathing or feeding. Depending on the symptoms observed, treatment might range from a change in position to tongue-lip adhesion, distraction osteogenesis of the mandible, or tracheostomy.

The first treatment choice should be position change.

Infants with less severe manifestations can be managed by placement in the prone position, taking advantage of gravity to draw the tongue forward. An oral airway or nasopharyngeal intubation often successfully relieves distress if the infant does not improve. However, in cases of persistent respiratory distress, cyanotic attacks while feeding, and failure to gain weight, surgical approach is necessary to relieve the obstruction.

When considering operative approaches, Schaefer et al.⁵ recommended starting with the least invasive treatment and then moving on to more invasive methods. In other words, when surgical relief is indicated, tongue-lip adhesion should be performed first before considering tracheostomy or distraction osteogenesis of the mandible.

The concept of tongue-lip adhesion for treatment of upper airway obstruction associated with Pierre Robin sequence was first proposed by Douglas in 1946.³ Tongue-lip adhesion consists of suturing a rectangular mucosal flap under the tongue to a mucosal flap of the lower lip with a traction suture from the tongue dorsum to the chin. It is a simple procedure with relatively rapid and significant clinical improvement after surgery. It is also a safe and effective surgical intervention in patients with Pierre Robin sequence and airway obstruction. Although it is a simple procedure, complications can still occur. It has been reported to be associated with postoperative complications, including dehiscence and injury to the adjacent structures.⁶ The tension of traction suture causes wound disruption of the flap of lower lip and invasion of traction suture through the alveolar ridge, resulting in dehiscence of tongue lip adhesion (Fig. 6). Wound dehiscence occurred in 17% to 42% of the patients.² Therefore Cauoette-Laberge et al.¹ gradually



Fig. 6. Traction suture invading alveolar ridge and causing injury can be seen in patients with dehiscence.

abandoned tongue-lip adhesion because of frequent dehiscence and the need for revisions.

During the infant period, there is a fibrocartilaginous union of the two halves of the mandible (Fig. 7), which becomes an osseous union during the first year. The symphysis menti is the line of junction of the two pieces of which the bone is composed at an early period in life. Resistance to traction of the tongue can be encountered with nonunionized symphysis menti, causing loosening of the traction suture through the symphysis menti (Fig. 8). This can lead to backward positioning of tongue, resulting in dehiscence of tongue lip adhesion.

To overcome the above limitations, an alveolar protector



Fig. 7. Three-dimensional CT image of a patient with Pierre Robin sequence. The fibrocartilaginous union of the two halves of the mandible is visible. It becomes an osseous union during the first year.

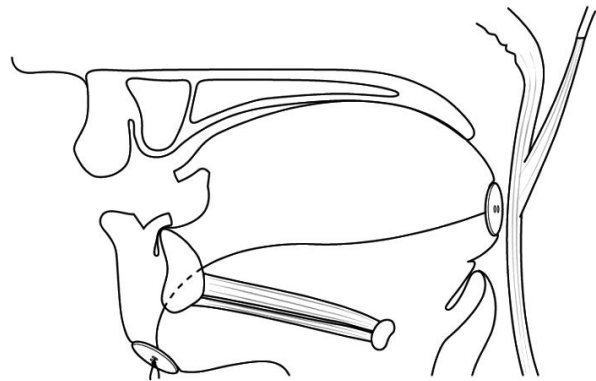


Fig. 8. Resistance to traction of the tongue is encountered with nonunionized symphysis menti. Tongue-lip adhesion can fail due to the suture loosening through the symphysis menti.

appliance can be used. This article describes the efficacy of the alveolar protector appliance for patients with Pierre Robin sequence. The alveolar protector appliance is a prosthesis that is designed to restore the separation between the ventral surface of the tongue and the lower labial sulcus in order to prevent the wound from being irritated and the suture from loosening (Fig. 4).

To prevent the protector from working as a space occupying material between lower labial sulcus and tongue tip to increase tension, we applied protector before adjusting tension. Also to prevent wound maceration and friction between the flap of the lower lip and the alveolar ridge, we used vinyl polysiloxane as the protector material with its high viscosity working as a buffer between the flap of the lower lip and the alveolar ridge. The putty type vinyl polysiloxane is the material of choice because its high viscosity reduces the danger of wound maceration. It can be effective in overcoming some of the wound problems associated with tongue-lip adhesion.

The traction vector can be more anterior to maintain the forward position of the tongue. It helps to place the tongue in the correct position of its functional role during treatment of the airway obstruction.

In the two tongue-lip adhesion procedures performed with alveolar protector, we observed no postoperative wound problems such as dehiscence. The time required to place the alveolar protector appliance is about 10 minutes, and we encountered no problems with the

operation. The alveolar protector appliance can be an effective method for securing tongue-lip adhesion. We believe that use of an alveolar protector appliance in tongue-lip adhesion can decrease the frequency of wound dehiscence. It is a simple and effective auxiliary method that yields functional improvement. Thanks to the aforementioned advantages, the alveolar protector appliance can be used widely for patients with the Pierre Robin sequence.

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