

A Robin sequence patient treated by the tongue-lip adhesion technique of modified Argamaso method

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[Received October 24, 2003 & Accepted November 25, 2003]

Abstract A 12-week-old boy with Robin sequence was referred to us for respiratory improvement by a pediatrician in Hachinohe Red Cross Hospital. The patient had micrognathia, upper airway obstruction, incomplete cleft palate and ankyloglossia. The patient had a history of repeated cyanotic episodes and had been under respiratory care with endotracheal intubation for more than 2 months. Using flexible fiber optic nasopharyngoscopy, his pharyngeal obstruction was classified as Type 1 of Sher's classification. Thus, we performed glossopexy of modified Argamaso method at 15 weeks after birth. The patient has had no cyanotic episodes at 2 years and 5 months postoperatively. We conclude that the glossopexy was effective for this patient.

Key words Robin sequence, cleft palate, ankyloglossia, airway obstruction, glossopexy

Introduction

For years Pierre Robin syndrome was known as the syndrome in which upper airway obstruction was observed in newborns with micrognathia and glossoptosis. However, this syndrome was associated with various conditions such as Stickler syndrome, velocardiofacial syndrome and Treacher Collins syndrome¹⁻³⁾. Micrognathia caused cleft palate by preventing the fusion

of the palatal processes in the prenatal period, and it led to upper airway obstruction in infancy as well. Thus, more recently the term "sequence" has been substituted, implying that one condition led to the next and then the next, and "Pierre Robin syndrome" is now called "Robin sequence"¹⁾.

Upper airway obstruction in the newborn causes cyanosis and/or pneumonia, and it causes the feeding problems of these

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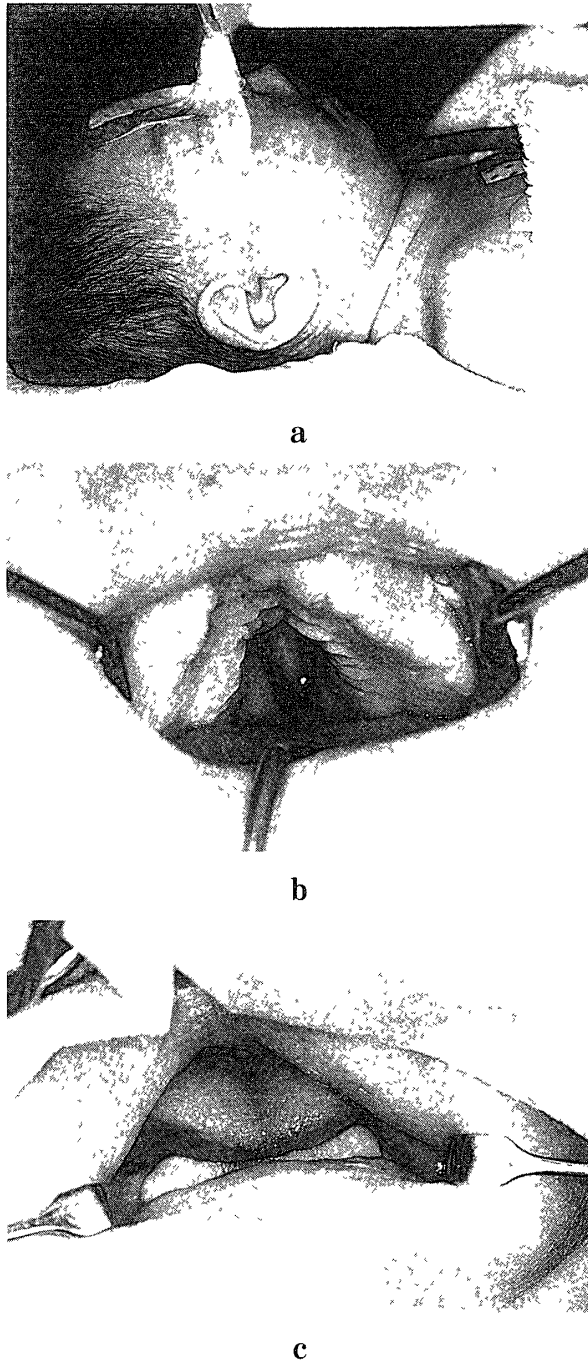


Fig 1 Preoperative findings from the glossopepy

a Lateral facial findings The patient showed micrognathia

b, c Intraoral findings The patient had incomplete cleft palate and ankyloglossia

children and their failure to gain weight, which often is serious and leads to death from athrepsia. Therefore, appropriate treatment is of vital importance for patients with Robin sequence.

Case Report

A 12-week-old boy with Robin sequence was referred to us for respiratory improvement by a pediatrician in Hachinohe Red Cross Hospital. The patient was born at 37 weeks' gestation after a normal delivery with micrognathia, upper airway obstruction and incomplete cleft palate, at a private practice. The birth weight was 1318 g. He was immediately referred to the pediatrics department of Hachinohe Red Cross Hospital and was hospitalized in the neonatal intensive care unit (NICU). He underwent respiratory care and nutrition improvement by the pediatric team. For the respiratory problem, he was treated with oxygenation and positioning for the first 3 weeks. However, endotracheal intubation was applied due to frequent severe airway obstruction. He was diagnosed as incomplete cleft palate and ankyloglossia at our first examination at 12 weeks after birth, and classified his pharyngeal obstruction as Type 1 of Sher's classification³⁾ by flexible fiber optic nasopharyngoscopy. Thus, the patient underwent the glossopepy with a modified Argamaso method²⁾ at 15 weeks after birth when his nutrition was improved.

Figure 1 shows the findings from the operation and Figure 2 shows the schemata of the incision design. A local anesthetic agent consisting of 0.5 percent Xylocaine with 1:200,000 dilution of epinephrine solution was infiltrated into the buccal surface of the lower lip, alveolar midline, anterior border of the tongue, and the tongue's undersurface. Two heavy sutures (2-0 silk) were placed on the bilateral sides of the tongue. These sutures were for protruding the tongue anteriorly. A

horizontal incision on the buccal surface of the lower lip was made from one commissure to the other with a #15 blade. The gingival mucosa was incised vertically

across the midline and the wound margins slightly undermined from the alveolus. The genioglossus muscle was gently detached by blunt dissection to gain tongue looseness. As the frenulum of the tongue was short, it was lengthened by simple transverse incision at the middle of lingual frenulum. The next incision was made on the undersurface of the tongue from its tip to the frenulum, and along the anterior border of the tongue (Fig. 2 a). An absorbable stay suture (4-0 Vicryl) was placed deeply into the tongue muscles and two absorbable sutures were placed through the base of the tongue to the surface. The stay sutures were looped around the mandible with an awl and stitched to the tongue muscle through the vertical incision on the tongue (Fig 2 b). As

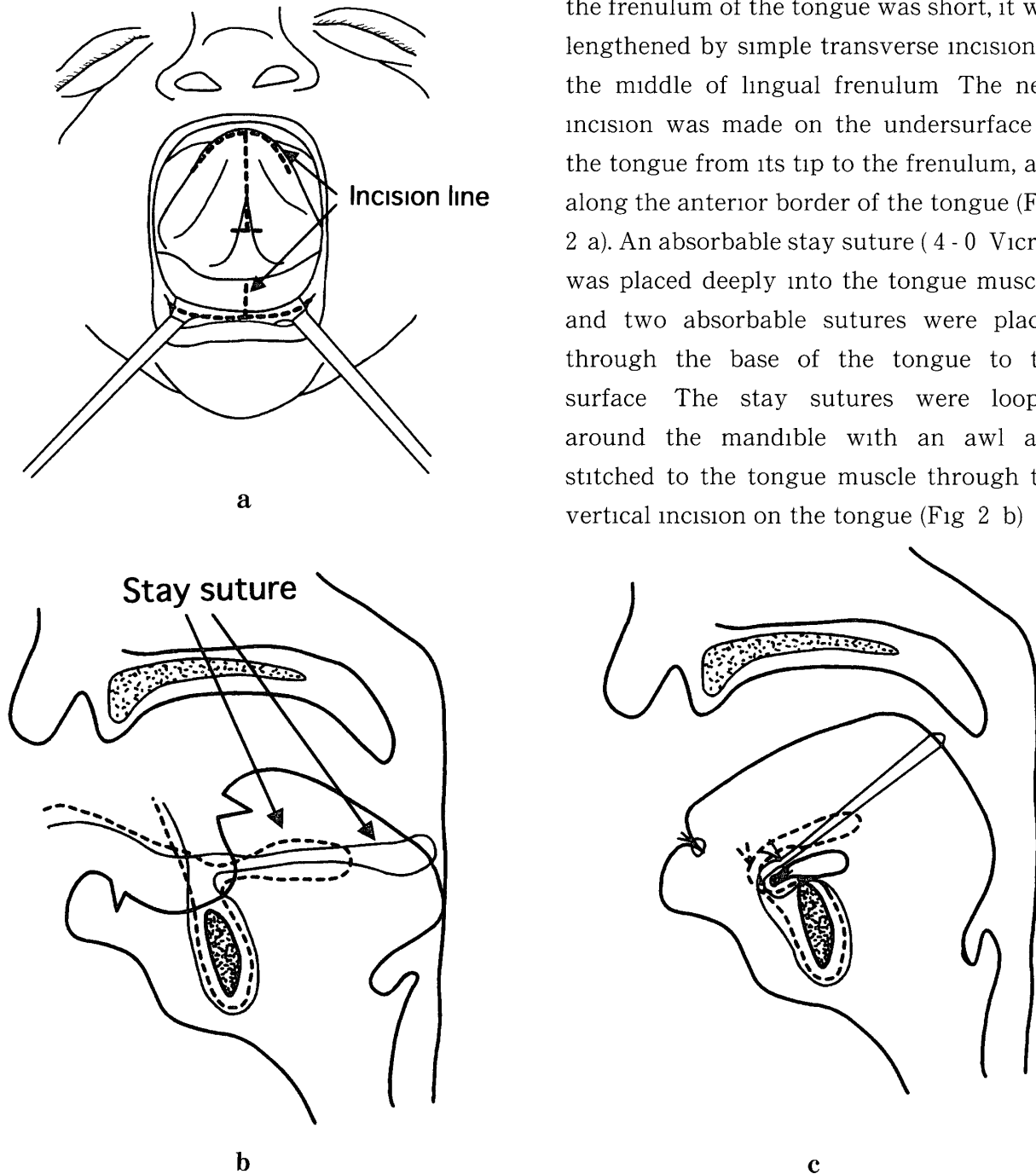


Fig 2 The schemata of the operation techniques

- a Incision design. The lines of incisions are marked along the anterior border of the tongue, with a vertical component from its midline to the undersurface, and along the buccal side of the lower lip, with a vertical component from its midline to the midline of the alveolar ridge.
- b Three absorbable stay sutures. An absorbable stay suture was placed deeply into the tongue muscles and two sutures were placed through the base of the tongue to the surface.
- c The completion of sutures. When these stay sutures were tied, the tongue was brought forward.

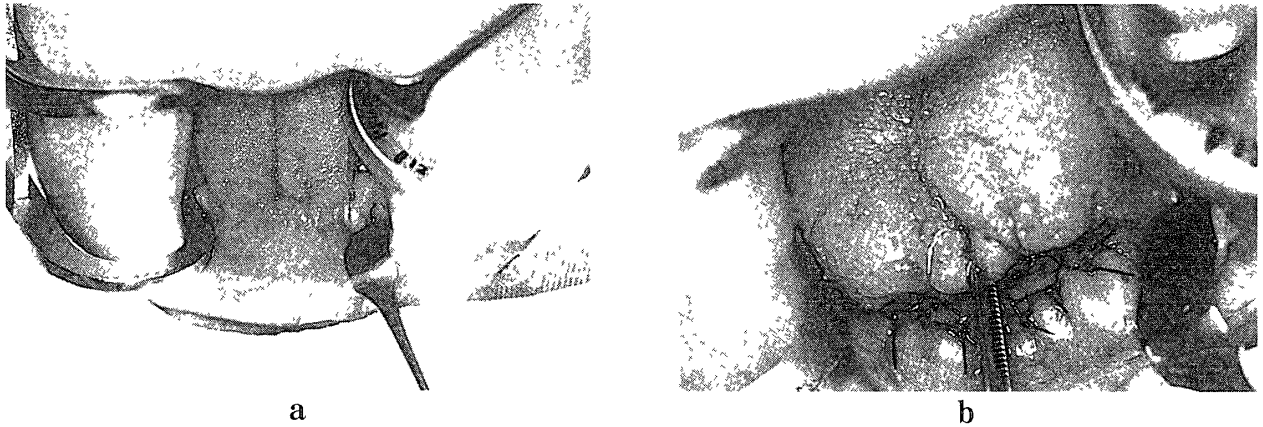


Fig 3 Intraoral findings at the tongue release
a Preoperative view Tongue-lip adhesion was complete
b Postoperative view The tongue and the lip were detached

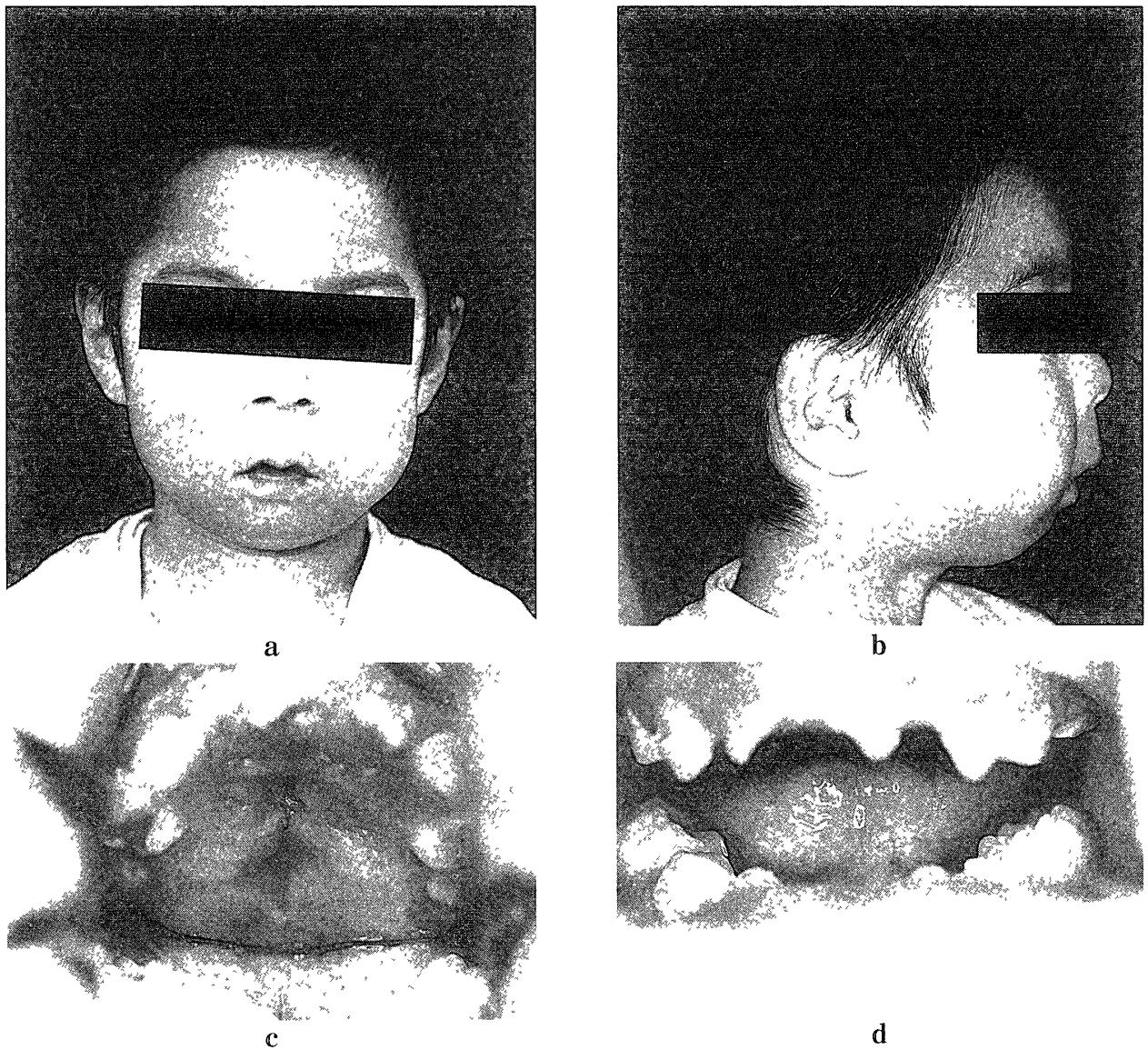


Fig 4 Findings at 2 years and 6 months of age
a, b Frontal and lateral facial views The patient has micrognathia
c Intraoral findings (palate) The cleft is closed completely
d Intraoral findings (lip and tongue) A slight scar is on the lip and tongue

these stay sutures were tied, the tongue was brought forward (Fig 2 c) At this point, the secureness of the upper airway was checked with nasopharyngoscopy With some undermining, the ventral tongue flaps were matched with the posterior flaps of the lower lip and sutured with 5-0 Vicryl Finally, the dorsal tongue flap was approximated to the anterior flaps of the lower lip Two heavy sutures on the bilateral sides of the tongue were left for emergency tongue traction

An endotracheal tube remained in place for 5 days The patient was observed carefully and was discharged 1 month postoperatively He withdrew from tube feeding 2 months postoperatively The tongue was released at the same time as palatoplasty at 18 months of age (Fig 3) The patient showed slight upper airway obstruction temporarily but no cyanotic attacks postoperatively Figure 4 shows the findings of the face and the oral cavity at 2 years and 9 months of age

Discussion

Many management methods for respiratory problems of Robin sequence have been reported^{1 2)} These are classified into nonsurgical management and surgical management In the nonsurgical management, the representative method is the positioning which keeps infants in a prone position in order to maximize the effects of gravity in keeping the tongue forward A variety of traction devices to draw the tongue or mandible forward have been reported⁴⁾ Even with all of the recommended treatments, the reported mortality rates are high²⁾ On the other hand, as to the surgical management, Shukowsky⁵⁾ reported the

instant relief from respiratory obstruction by suturing the tongue to the lip Douglas⁶⁾ popularized the concept of the lip-tongue adhesion Other techniques have been devised to fix the tongue to the cheeks⁷⁾ or the mandible⁸⁾ Furthermore, Hadley⁹⁾ reported Kirschner wire could be placed through the angles of the mandible, transfixing the base of the tongue in a forward position

As to the indication of surgical treatment, Parsons and Smith reported that surgery should be considered in the case that after seven days of conservative management the child was still unable to control the airway and did not gain weight, and in the case that the child was still dependent on an endotracheal tube for an adequate airway after three days of intubation^{1 10)} Sher et al³⁾ utilized flexible fiber optic nasopharyngoscopy for the surgical indication According to their report, the Type 1 obstruction consisted of the posterior movement of the dorsum of the tongue to the posterior pharyngeal wall so that the majority of airway obstruction was anteroposterior, and that this type responded well to glossopexy³⁾

Our patient was treated with oxygenation and positioning for the first 3 weeks and endotracheal intubation for more than 2 months However, he had a history of repeated cyanotic episodes, and thus, we decided on surgical treatment We chose the lip-tongue adhesion method by nasopharyngoscopic findings which showed Sher's Type 1 obstruction

Several techniques of glossopexy have been reported The Douglas method consists of denuding a rectangular area under the tongue, along the floor of the mouth, over

the alveolus, and onto the lower lip⁶⁾ However, in this method, surgeons experienced complications including tongue lacerations, wound infections, dehiscences, injuries to Wharton's ducts, and scar deformations of the lip, chin, and floor of the mouth Routledge¹¹⁾ described a simple method that provided a wider surface contact for adhesion between the tongue and lip, to avoid scarification of the floor of the mouth In the Routledge method, a generous incision was made along the length of the lower lip and a matching incision was made around the anterior edge of the tongue Parsons and Smith¹⁰⁾ stressed the importance of releasing the genioglossus from the symphysis of the mandible Argamaso²⁾ reported the operation technique using two points of attachment for the tongue, one at the mandibular alveolus and the other at the lower lip by two T-shape incisions on the anterior tongue and on the lower lip and alveolus He also stressed that the genioglossus should be released to lengthen the tongue and emphasized the importance of the stay suture looped around the mandible and stitched to the tongue muscle

We performed the glossopexy using a modified Argamaso method Our modified points were as follows

- (1) We placed three absorbable buried stay sutures for drawing the tongue to the mandible, and two of these sutures were placed on the surface of the base of tongue In the original technique, only two stay sutures were placed into the lingual muscle Three stay sutures made tongue fixation more secure
- (2) We placed two heavy sutures on the bilateral sides of the tongue to protrude the

tongue anteriorly, and left the sutures for postoperative emergency

- (3) We lengthened the frenulum simultaneously at the time of T-shape incision on the undersurface of the tongue

The patient is 2 years and 9 months of age and has had no cyanotic episodes postoperatively We conclude that the glossopexy was effective for this patient However, he still has micrognathia, so we are planning to perform distraction osteogenesis of the mandible in the future for this problem

Acknowledgements

We would like to extend our special thanks to the patient's family for accepting the publication of this report We also wish to thank the staff of the Pediatrics Department and the Dentistry, Oral and Maxillofacial Surgery Department of Hachinohe Red Cross Hospital, and the Second Department of Oral and Maxillofacial Surgery of Iwate Medical University for their help in this report

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Argamaso 変法による舌固定術を行った Robin sequence の 1 例

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(受付 2003年10月24日)

(受理 2003年11月25日)

抄録 われわれは八戸赤十字病院小児科から, *Robin sequence* の生後12週男児の呼吸障害の改善のための対診を依頼された。患者は下顎後退, 上気道閉塞, 軟口蓋裂, 舌小帯強直症がみられた。当科を受診前に, 何度か重度の呼吸障害がみられ, 2か月以上気管内挿管による呼吸管理が行われていた。患者の咽頭部の閉塞は, 内視鏡検査による *Sher* の分類の *Type 1* であったため, われわれは生後15週で *Argamaso* 変法による舌固定術を行った。術後2年5か月経過するか, 以来患者にチアノーゼは見られず, 舌固定術は同患者に有効な治療法と思われた。

キーワード Robin sequence, 口蓋裂, 舌小帯強直症, 気道閉塞, 舌固定術