The Elongation of Distal Esophageal Pouch by Mechanical Bougienage for a Year Resulted in a Tension-free Anastomosis in a Patient with Long Gap Esophageal Atresia: A Case Report

SHIGEKI HIKIDA, YOSHIKAI TANAKA, TOMOMITSU TSURU, KENJIRO AKIYOSHI, KIMIO ASAGIRI, HIROTAKA NAKAMIZO, HIROKO SOEJIMA, MARI OHTANI, SUGURU FUKAHORI, HIDEFUMI KOBAYASHI, AKIKO KAIDA, TAKAHIRO ASAGAWA AND HIROYOSHI MIZOTE

Department of Pediatric Surgery, Kurume University School of Medicine, Kurume 830-0011, Japan

Summary: Despite the numerous approaches described for the management of neonates with “long gap” esophageal atresia, controversy still exists as to the preferred method. Delayed primary anastomosis is probably the most frequently adopted practice but often the native esophagus is abandoned. We report a case of a 2.98 kg newborn with pure esophageal atresia. Although the elongation of the distal esophageal pouch by mechanical bougienage was initiated at 1 year and 8 months, a successful tension-free anastomosis with minimum dissection of the lower esophagus was performed at 2 years and 7 months. Her postoperative quality of life has been quite excellent. This report emphasizes that a tension-free anastomosis without operating on the lower esophagus and stomach is essential for the treatment of long-gap esophageal atresia.

Key words long-gap esophageal atresia, tension-free anastomosis, bougienage

INTRODUCTION

Modern management of the newborn with esophageal atresia has enabled the survival rate of infants with this condition to approach 100% with minimal postoperative complications [1]. However, the difficulty in achieving a primary anastomosis increases proportionately with the length of the gap between the proximal and distal ends of the esophagus. In infants with pure esophageal atresia, the esophageal segments are usually too far apart to enable a primary anastomosis to be performed. Current techniques to gain extra length are prone to complications and esophageal replacement remains a poor long-term substitute for the patient’s own esophagus. This report describes the usefulness of preoperative mechanical bougienage for elongation of the distal esophagus to establish tension-free esophageal continuity in long-gap esophageal atresia utilizing the native esophagus.

CASE REPORT

A 2980 g girl was born at 38 weeks gestation. A nasogastric tube could not be passed into the stomach and an abdominal X-ray showed the nasogastric tube coiled in a dilated upper esophageal pouch together with a gasless abdomen. A diagnosis of pure esophageal atresia was made with the expectation of the presence of a long gap. Echocardiography and laboratory examination showed that she also had an atrial septal defect and congenital hypothyroidism. Downs syndrome was diagnosed by chromosomal examination.

She was fed by gastrostomy and continuous suction was applied to the upper pouch to aspirate her saliva. Although continuous suction ensured that she...
HIKIDA ET AL. did not develop any episodes of aspiration pneumonia while she was in the previous hospital, her nutritional status remained poor and her congenital hypothyroidism was not well controlled. When she was 1 year and 8 months old, she was admitted to our hospital for additional nutritional management and definitive treatment of the esophageal atresia. On admission, her body weight was 4176 g (less than -2SD) and her height was 60.3 cm (less than -2SD). She received aggressive nutritional management and treatment for her congenital hypothyroidism was instituted by a pediatric endocrinologist. Her atrial septal defect had spontaneously closed. Continuous suction of the upper esophageal pouch to deal with her saliva was maintained. Regarding the esophageal atresia, her esophageal gap was greater than 2 vertebral bodies (Fig. 1) on admission. Therefore, mechanical bougienage of the distal esophagus was performed through a gastrostomy using a size 8 Hegar dilator. The bougienage was performed 20 times as a total. Eventually, the gap was shortened to less than half of a vertebral body (Fig. 2) when she was 2 years and 7 months old. Her body weight had reached 10.2 kg (less than -2SD) and her height had reached 74 cm (less than -2SD). Her thyroid function was well controlled with appropriate medication.

Radical surgery for the esophageal atresia was performed. The child was placed in the left lateral position and a muscle-sparing thoracotomy was performed via the fifth intercostal space. The proximal esophageal pouch was identified and dissected off the trachea as proximally as technically feasible. The size 8 Hegar dilator was inserted to the distal esophagus via a gastrostomy to identify the distal esophageal pouch which was dissected above the azygos vein. The two ends of the esophagus were opened and a primary, single-layer end-to-end anastomosis was performed using 4-0 absorbable monofilament. No additional maneuver was necessary since the anastomosis was completely free of tension and the delicate blood supply of the distal esophageal pouch was evident following completion of the anastomosis.

An intercostal catheter was placed adjacent to the esophageal anastomosis and the intubated and paralyzed patient was returned to the intensive care department. A wound infection developed but resolved after open drainage was performed.

A water-soluble contrast study showed no evidence of anastomotic leak but a slight stenosis was apparent. As a result, she required bougienage using a size 24 mercury dilator on 2 occasions during the subsequent 3 months. The esophagogram 3 months after operation exhibited no evidence of gastro-esophageal reflux or a hiatus hernia. In order to enable weight gain she was maintained on gastrostomy feeding for several weeks whilst introducing oral feeding. Subsequently, she had no problems with swallowing and her oral intake increased. The

![Fig. 1. On admission, the anteroposterior projection indicates the initial gap of more than 2 vertebral bodies between the esophageal segments in the patient (aged 1 year and 8 months).](image1)

![Fig. 2. Anteroposterior view at 2 years and 8 months demonstrates marked shortening of the gap to less than half of a vertebral body.](image2)
gastrostomy was removed 5 months following surgery.

DISCUSSION

The fact that the number and the variety of procedures have been described for the management of long gap esophageal atresia is a testament to the difficulties that this condition continues to provide for pediatric surgeons worldwide. The most common method of managing such infants with preservation of the host esophagus is by performing a delayed primary anastomosis and this strategy was adopted in this case. Using this method, the ideal time for performing a delayed primary anastomosis is when the infant is 8 to 10 weeks old [2] since the maximum natural growth of the end of the esophagus is seen in the first 2 to 3 months [3]. In this case, her age was 1 year and 8 months on admission but her body weight was 4176 g which is approximately the weight of a normal 5 week old baby. We judged that she had not received adequate nutrition to facilitate her normal systemic growth and development including the esophagus. In addition, her nutritional status together with inadequate hormonal control of her hypothyroidism was too poor to facilitate immediate surgery for the long-gap esophageal atresia. Fortunately, despite the long duration of continuous upper esophageal suction in the previous hospital, she had not developed the dangerous complications of aspiration pneumonia and her history did not suggest the presence of a proximal fistula in her upper esophageal pouch. Thus it was considered that she could tolerate the continuous suction method and a decision was made to perform a primary delayed anastomosis.

Regarding the esophageal elongation, a variety of adjuvant methods have been described to promote interim esophageal growth. These include mechanical bouginage of the upper pouch [4] and/or distal pouch (via the gastrostomy [5]) or the use of mercury filled bags [6] and magnets [7]. We usually use mechanical bouginage of the distal pouch via the gastrostomy and we have not experienced perforation from this treatment. We applied this method to this case twice a week and the gap became markedly shortened after a year.

In many cases which undergo primary delayed anastomosis, the anastomosis is still completed under considerable tension. A variety of tension-relieving procedures have thus been used in an attempt to minimize this. A circular myotomy of the proximal pouch (Livaditis) [8,9] has been associated with complications [10] that Kimura et al. [11] attempted to minimize by utilizing a “spiral” myotomy and burying the proximal pouch subcutaneously in preparation for a delayed second reconstruction. Finally, a tube fashioned from a full-thickness proximal pouch flap has been used [12], but this method has been associated with significant complications and uncertain long-term results [13].

In some centers, primary esophageal replacement is the preferred method for patients with long-gap esophageal atresia. Various segments of the gastrointestinal tract have been utilized for esophageal substitution and these have been variably placed via subcutaneous, retrosternal, transthoracic and posterior mediastinal routes. Methods utilizing the stomach include creating a “tube” from the greater or lesser curve [14], total gastric transposition as advocated by Spitz [15], division of the lesser curvature as described by Scharli [16] and use of a Collis gastroplasty [17]. The small intestine has also been utilized. However, the colon placed in an iso- or antiperistaltic direction is currently the most common choice for esophageal reconstruction [18]. The successful completion of esophageal continuity utilizing any of the previously described techniques is initially well tolerated. However, all of these techniques are associated with significant long-term complications [19-22].

Basic tenets of surgery include the avoidance of tension and preservation of the blood supply of the anastomosis. These are inevitable in any early or delayed repair of long gap esophageal atresia. In our patient, mobilization of the distal esophageal pouch below the azygos vein was not necessary in order to perform a primary anastomosis. Therefore, the segmental blood supply to the lower esophageal pouch from the aorta was well preserved. In addition, the physiological and anatomical status of the lower esophagus and stomach was well preserved and these are essential for the prevention of gastro-esophageal reflux. Although she needed postoperative bouginage twice during the postoperative 3 months, the physiologically and anatomically normal lower esophagus and stomach together with strong familial and experienced nursing support and care contributed to her increased oral intake.

In conclusion, persistent, gentle and long-term bouginage of the distal esophageal pouch contributes to the elongation of the distal esophagus. This maneuver results in a tension-free anastomosis with minimum dissection and this is beneficial for the postoperative quality of life in patients with long-gap
esophageal atresia.

REFERENCES


