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Outcomes of primary gastric transposition for long-gap esophageal atresia in neonates

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Abstract

Background: Gastric transposition is a relatively novel method of esophageal replacement. The purpose of this retrospective study was to assess the outcomes of long-gap esophageal atresia (LGEA) treated with esophageal replacement using primary gastric transposition in neonates.

Methods: Between March 2008 and May 2015, 14 newborns with LGEA were treated in our hospital. They were all found to have gaps of over 3 cm at the time of the surgery and were diagnosed with LGEA. Primary gastric transposition was performed. They also underwent a gastric drainage procedure by pyloromyotomy. The nasogastric tube was removed if no anastomotic fistula was present and oral feeding was initiated. After initial recovery and discharge, the patients were evaluated with outpatient follow-ups or telephone follow-ups from 1 month after the surgery.

Results: The mean age of the neonates at the time of the surgery was 32 hours (range, 4–96 h). The mean birth weight was 2550 g (range, 2100–3500 g). There were 2 deaths in this series of patients due to respiratory failure or withdrawal of treatment by the parents, with a mortality rate of 14.3%. Seven of the neonates developed unilateral or bilateral severe pneumonia. Early anastomotic leak occurred in 3 cases and anastomotic strictures occurred in 4 cases. These 4 neonates were able to eat a fairly normal diet after esophageal balloon dilation. Gastroesophageal reflux occurred in 7 of 12 cases. Feeding multiple small meals and postural support for positioning and feeding were instructed for these 7 cases. Subsequently, the symptoms alleviated and they had no additional surgical therapy. None of the neonates had delayed gastric emptying or gastric retention.

Conclusion: Primary gastric transposition may be a rewarding reconstructive option in the treatment of LGEA.

Abbreviations: EA = esophageal atresia, GER = gastroesophageal reflux, ICU = intensive care unit, LGEA = long-gap esophageal atresia.

Keywords: anastomotic leak, anastomotic strictures, long-gap esophageal atresia, primary gastric transposition, pyloromyotomy

KEY POINTS

1. A retrospective study was performed on 14 newborns with long-gap esophageal atresia.
2. Low incidence of anastomotic leaks and strictures was identified.
3. All the infants had no delayed gastric emptying or gastric retention.

1. Introduction

Esophageal atresia (EA), with or without tracheoesophageal fistula, is a congenital malformation of the esophagus.[1] The incidence of EA is approximately 1 in 3000 live births.[2] To divide and ligate the fistula and anastomose the esophageal segments is imperative as soon as possible after birth.[3] The survival rate of newborns with EA has significantly increased during the last few decades.[4] However, it continues to be a challenging problem to identify an ideal management protocol for these infants because the clinical management may be fraught with postoperative complications, such as chronic recalcitrant strictures and anastomotic leaks.[5]

Brown and Tam[6] proposed a classification based on the length of the gap between the esophageal segments (long-gap: >3 cm; intermediate-gap: >1 cm but ≤3 cm: and short-gap: ≤1 cm) to address the magnitude of the surgical problems in EA and tracheoesophageal fistula. They found that this classification could predict morbidity and long-term outcomes associated with EA surgeries, and long-gap EA (LGEA) was associated with poor outcomes.[6] Ideal surgical treatment of EA includes division of the tracheoesophageal fistula as well as a primary end-to-end anastomosis of the upper and lower esophageal segments. However, the outcome of this approach may vary based on the presence of pulmonary complications or related cardiac congenital anomalies.[7] LGEA further complicates the debate on EA and is still a major challenge.[8] Failure to achieve a satisfactory primary esophageal anastomosis will require esophageal replacement with the stomach,

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colon, or small intestine.[8] Controversy remains regarding the many options available for the surgical management of LGEA.

Gastric transposition is a relatively novel method of esophageal replacement.[9] We have favored the gastric transposition as a procedure of choice for the treatment of LGEA in our institution. This study was to retrospectively review our surgical experience and assess the outcomes of LGEA treated with esophageal replacement using primary gastric transposition in neonates.

2. Materials and methods

2.1. Patients

Between March 2008 and May 2015, 14 newborns (11 male and 3 female neonates) with LGEA were treated in our hospital. Furthermore, 2 of them were premature infants. EA was diagnosed by iodized oil radiography of the esophagus and plain abdominal radiographs. The cases were classified into 2 Type I and 12 Type IIIa of the Gross classification.[10] All patients were found to have gaps between the esophageal segments of over 3 cm at the time of the surgery and were diagnosed with LGEA. Three cases were complicated by ventricular septal defects and 8 cases had atrial septal defects. Patent ductus arteriosus, thumb polydactyly, and left hydronephrosis were found in 1 case each. One case was complicated by, both, ventricular septal defect and atrial septal defect. One case had complicated atrial septal defect and left hydronephrosis. Primary gastric transposition was performed in all the 14 neonates. This study was approved by the institutional ethics committee, and written informed consent was obtained from the parents of all the neonates.

2.2. Surgical approach

The neonates underwent general anesthesia and endotracheal intubation, and were then placed in the left lateral position. The surgical technique used has been previously described with modifications.[11,12] Posterolateral incision was made at the 4th intercostal space at the right side. We measured the distance between upper and lower esophageal pouch. If the distance was over 3 cm, esophageal end-to-end anastomosis could not be performed and, therefore, gastric transposition was carried out. The proximal esophageal caecum was fully dissected till the bottom of the pharynx. Subsequently, the tracheoesophageal fistula was ligated, and the distal esophagus was bluntly dissected up to the level of the diaphragm. Then the surgery in the abdomen was performed. The abdomen was accessed through a left subcostal incision. The left gastric artery and vein, as well as the short gastric vessels, were dissected and ligated. The right gastric artery was identified and preserved. All the neonates underwent an associated gastric drainage procedure by pyloromyotomy. Part of the stomach was pulled up into the chest and the residual fraction of the distal esophagus was preserved. An anastomosis was carried out between the apex of the fundus and the distal cervical esophagus. A chest drainage tube was retained. A 6 French nasogastric tube was placed during anastomosis in order to provide postoperative enteral nutrition.

2.3. Postoperative monitoring and follow-up

After the surgery, the neonates were admitted to the intensive care unit (ICU), with assisted respiration by a respirator. We turned over the infants and patted on their backs on occasion. The infants were maintaining the fluency of the respiratory tract and prophhylaxis against pulmonary infection was provided by using the appropriate antibiotics based on the results of sputum bacteria culture. On day 1 postoperatively, they were provided with total parenteral nutrition. After 2 to 3 days, glucose in water was administered through the nasogastric tube with micro pump for 1 to 2 days and, subsequently, appropriate amounts of milk were administered. Esophageal radiography examination was performed at 1 week postoperatively. The nasogastric tube was removed if no anastomotic fistulae were present and oral feeding was initiated. For neonates with anastomotic fistulae, feeding was continued with the nasogastric tube.

After initial recovery and discharge, the patients were followed up by outpatient consultation or telephone calls, starting 1 month after surgery. The follow-ups were carried out at monthly intervals for the first 6 months. After 6 months, follow-up was carried out semiannually. Radiological esophageal imaging was performed at 2 months and 1 year after the surgery. Additionally, we performed respiratory function tests for 6 children after 3 years of follow-up. Children ≤ 5 years of age who undergo such surgeries generally cannot meet the traditional pulmonary function testing. For 2 to 5-year-old children with lung function tests, sleep state may serve as a diagnostic window.[13] Children who cannot fall asleep were treated with drug-assistance. Tidal volume, respiratory rate, inspiratory time, expiratory time,[14] the ratio of the proportion of time to reach peak tidal expiratory flow to total expiratory time (tPTEF/tE),[15] and the ratio of volume until peak expiratory flow volume to total expiratory volume (vPTEF/vE)[16] were evaluated. Through the above tests, comprehensive evaluation of lung functions of the children was performed. Additionally, during follow-up, the infants were observed whether recurrent vomiting and regurgitation phenomenon occurred; if the infants had normal sucking without vomiting/regurgitation, they were determined to have no delayed gastric emptying or gastric retention. For the childhood, they were observed whether recurrent vomiting and upper abdominal satiety occurred; if they had normal eating without recurrent vomiting and stomach discomfort, they were determined to have no delayed gastric emptying or gastric retention. For the children with suspected delayed gastric emptying or gastric retention, further evaluation was performed using color Doppler ultrasound.

3. Results

The mean age of the 14 newborns at the time of the surgery was 32 hours (range, 4–96 h). The mean birth weight was 2550 g (range, 2100–3500 g). There were 2 deaths in this series of patients (mortality rate 14.3%). One child died on the 7th postoperative day due to severe pneumonia with concurrent pyopneumothorax and respiratory failure. Another child died on the 5th postoperative day due to anastomotic fistula and withdrawal of treatment by the parents. Seven infants had unilateral or bilateral severe pneumonia; 1 infant developed concurrent pyopneumothorax and died, and pneumonia in the other 6 infants was treated by comprehensive treatment including assisted respiration, anti-infective therapy, and respiratory physiotherapy. Early anastomotic leak occurred in 3 neonates; parents of 1 neonate withdrew from the treatment and the neonate died, and the anastomotic leak was closed in the other 2 neonates through therapy such as milk via the nasogastric tube, parenteral nutrition enhancement, and anti-infective therapy.

In the 12 remaining cases that were discharged from the hospital, follow-up was carried out for 1–6 years. Figure 1 shows

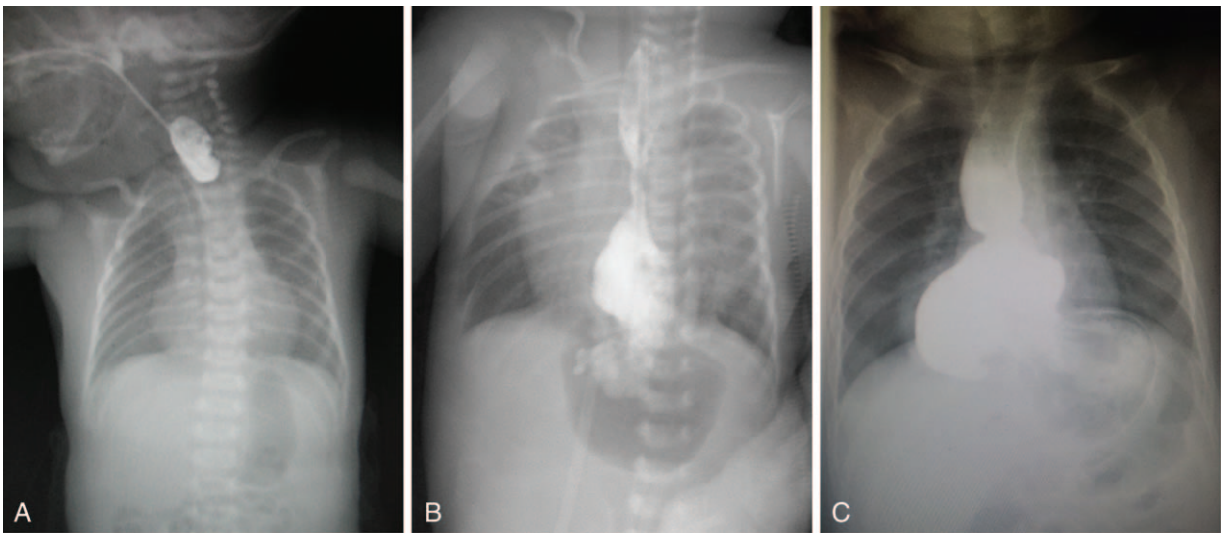

Figure 1. Results of radiography before the surgery and at 2 months and 1-year post-operatively of case number 9, respectively. (A) Radiography before the surgery. (B) Radiography at 2 months after the surgery. (C) Radiography at 1 year postoperatively.

the results of radiography before the surgery, and at 2 months and 1 year post-operatively of a case. As shown in Table 1, anastomotic strictures occurred in 4 cases. These 4 patients were able to eat a fairly normal diet after esophageal balloon dilation. Gastroesophageal reflux (GER) occurred in 7 of 12 cases. Feeding multiple small meals and postural support for positioning and feeding were performed for these 7 cases. The symptoms alleviated and they had no need for additional medical or surgical therapy. All the infants had no delayed gastric emptying or gastric retention. Body weight and height of 11 cases were normal and comparable to that of the children of the same age. The body weight of 1 child was less than his peers due to concomitant congenital heart disease and recurrent respiratory tract infections.

Additionally, because the parents of some of the infants did not agree with pulmonary function tests and some infants were too young for pulmonary function testing, we performed pulmonary function tests in only 6 children in 3 years of follow-up. We found that their vital capacity was less than that of their peers. However, no dyspnea, chest discomfort, and labored breathing were identified and the low vital capacity had no significant effect on the growth and development of the children.

4. Discussion

The surgical management of patients with LGEA remains controversial. Evidence shows that a tension-free primary esophageal repair or replacement that use alternative conduits can lead to relatively less dysfunction in motility in patients with EA undergoing this option compared with other procedures.[17] Methods employed for esophageal replacement consist of esophagocoloplasty, gastric tube interposition, small intestine interposition, and gastric transposition.[18–20] Esophageal replacement techniques have been found to have a number of associated complications such as anastomotic stricture, anastomotic leak, and reflux.[17] The ideal substitute of choice continues to be debated. Recently, Tannuri et al[21] showed that gastric transposition was preferable to gastric tube reconstruction.

Table 1

Postoperative complications and the therapeutic outcome of the included children.

Case	Complications	Outcome
Number 1	Severe pneumonia, anastomotic stricture, gastroesophageal reflux	Cured and left the hospital
Number 2	Severe pneumonia	Died
Number 3	Anastomotic fistula	Abandoning therapy and died
Number 4	Severe pneumonia, gastroesophageal reflux	Cured and left the hospital
Number 5	Anastomotic fistula, anastomotic stricture	Cured and left the hospital
Number 6	Anastomotic stricture, gastroesophageal reflux	Cured and left the hospital
Number 7	Severe pneumonia, gastroesophageal reflux	Cured and left the hospital
Number 8	Severe pneumonia, gastroesophageal reflux, recurrent respiratory infection	Cured and left the hospital; 20–30% weight behind their peers
Number 9	Severe pneumonia	Cured and left the hospital
Number 10	Gastroesophageal reflux	Cured and left the hospital
Number 11	None	Cured and left the hospital
Number 12	Gastroesophageal reflux	Cured and left the hospital
Number 13	Anastomotic fistula, severe pneumonia, anastomotic stricture	Cured and left the hospital
Number 14	None	Cured and left the hospital

Additionally, Macksood et al[22] identified that the use of gastric transposition for management of LEGA in children had fewer complications and was relatively safer in comparison with another procedure. Gupta et al[9] performed gastric transposition for 27 neonates with EA and demonstrated that gastric transposition could be a lifesaving alternative even in the critically ill neonates with tracheoesophageal fistulae and leaks. These studies may provide clinical basis for the use of primary gastric transposition in LGEA treatment in neonates.

GER, anastomotic stricture, and anastomotic leak are common complaints in children with LGEA who were treated by delayed primary anastomosis.[23,24] Anastomotic leak can be devastating; it may lead to mortality as a result of irreversible sepsis and mediastinitis. A possible contributing factor to a leak is tension on the suture lines.[25] Besides, anastomotic stricture is the most common cause of revision surgery in these patients.[25] The advantages of gastric transposition are the requirement of a single anastomosis, excellent blood supply of the stomach, technical ease of the procedure, and the fact that adequate length is available for anastomosis,[11] leading to the lower incidence of anastomotic leak and stricture in gastric transposition compared with other procedures.[12] In this study, anastomotic strictures occurred in 4 cases. These 4 patients were able to consume a fairly normal diet after esophageal balloon dilation. GER is a common problem after gastric transposition.[26] In this study, 7 neonates would spit up milk after feeding. Conservative treatment, including multiple small meals and postural support for positioning and feeding, was performed for these neonates. They received multiple small meals and postural support for feeding in a right supine position with the head end elevated during infancy. During later childhood, they received multiple small meals and thickened diet. GER symptoms alleviated with age and they had no additional need for medical or surgical therapy. On the other hand, it is recommended to perform respiratory function tests after surgery.[27] In this study, we have performed respiratory function tests in 6 children during 3 years of follow-up, and we found that their vital capacity was lower than that of their peers. However, no dyspnea, chest discomfort, and labored breathing were identified and the low vital capacity had no effect on their growth and development.

Regarding the transposed stomach, studies have been performed on the necessity for a drainage procedure, including pyloromyotomy or pyloroplasty.[11,28] Compared with pyloroplasty, pyloromyotomy is easier to perform and incurs less damage to the gastric wall. A previous study achieved good results with a pyloromyotomy alone.[28] In the present study, all neonates underwent an associated gastric drainage procedure by pyloromyotomy. As a result, none of them developed delayed gastric emptying or gastric retention. On the other hand, a nasogastric tube was placed for all the neonates intraoperatively in order to provide early postoperative enteral nutrition which can reduce the use of parenteral nutrition, thus, reducing the incidence of parenteral nutrition-related complications.[29] Additionally, 2 cases with anastomotic leaks in this study were provided conservative management with enteral nutrition using nasogastric tube, which played an important role in promoting the natural healing of the leaks.

Primary gastric transposition used in this study may have improved the survival of infants with LGEA to a certain extent by reducing the incidence of postoperative complications. However, this open surgical approach requires a thoracoabdominal incision that may cause large trauma, and is more demanding of the anesthesiologist and surgeon involved. More studies are required before primary gastric transposition may be recommended. Thus, we recommend that those hospitals with extensive experience in EA treatment and with specialized pediatric centers could perform such surgery. Furthermore, regarding the use of ultrasonography for the diagnosis, as in this study, ultrasonography should be performed by an expert with paramount training. Several factors may have influenced the outcomes of the study, such as the selection of a single or multistage process by the parents of the infants, the technical expertise of the surgeon, and the severity of the associated malformations.

Our report has some limitations. First, the number of cases was small, which ruled out a case-control study design that would require a larger sample size. Nevertheless, we have referred to the relevant literature in the use of gastric transposition for the treatment of LGEA, and we selected primary gastric transposition for LGEA treatment in this study. The outcomes indicated that primary gastric transposition could provide a means of treatment to improve the cure rate of LGEA. Secondly, the retrospective nature of this investigation could impact the validity of the data.

In summary, we found that the outcomes of primary gastric transposition for LGEA are promising. Primary gastric transposition shortens the period of clinical treatment, reduces the economic and psychological burden on the parents of the children, thus improving the cure rate of LGEA to a certain extent. Thus, primary gastric transposition may be a rewarding reconstructive surgical option in the treatment of LGEA. However, there is a clear need for high quality randomized and comparative studies to establish recommendations and guidelines.

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