

Esophageal Replacement in Children: A 10-Year, Single-Center Experience

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ABSTRACT

Objective: Various esophageal replacement grafts have been used in children, although none can equal the native esophagus. The purpose of this study was to review the complications and outcomes associated with using different techniques in a single institute.

Methods: A retrospective medical record review was conducted from 2006 to 2016. Patient demographics, perioperative clinical courses, complications and long-term outcomes were reported as percentages and categorized according to the surgical procedure performed.

Results: A total of 15 children underwent esophageal replacement procedures, comprising 7 (47%) isoperistaltic gastric tubes, 3 (20%) colonic interpositions, 3 (20%) gastric transpositions and 2 (13%) reversed gastric tubes. Indications for esophageal replacement included long-gap esophageal atresia (5; 33%), esophageal atresia with severe postoperative complications (6; 40%), and caustic injury (4; 27%). The mean age of patients was 2.9 years (range: 0.2–15 years). The average follow-up duration was 3.6 years (range: 0.4–8 years). There was no perioperative mortality and no graft loss in any group. The long-term outcomes were acceptable, with no late stricture. Eighty-six percent of the patients in the isoperistaltic gastric tube group and all patients in the other procedural groups achieved full oral feeding. Nevertheless, the patients had various degrees of malnutrition.

Conclusion: Esophageal replacement remains a major challenge in children. Our experience indicates that children can be safely operated on using any of these methods, with acceptable outcomes and no deaths. Nevertheless, the long-term consequences and complications should be monitored throughout adulthood.

Keywords: Esophageal replacement; isoperistaltic gastric tube; reversed gastric tube; gastric transposition; colonic interposition (Siriraj Med J 2019;71: 80-88)

INTRODUCTION

Over the last several decades, many different techniques and various visceral substitute grafts have been used for esophageal replacement in children.¹⁻⁴ Common indications for the esophageal replacement procedure in children include long-gap esophageal atresia, severe peptic or caustic injuries, and anastomotic strictures.⁵⁻⁷

The ideal esophageal substitute should closely imitate the native esophagus both in size and function; nevertheless, none can match a normal esophagus.^{1,5} Several studies have reported comparable outcomes for each technique, with no significant differences in terms of their early and late complications.¹⁻⁷ The procedure selection and graft choice in those studies were based on the anatomy and

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availability of the visceral substitute and, in particular, the experience and preferences of the surgeon. Factors influencing the outcomes were related to the infrequency with which the procedures were performed, the variable expertise among the surgeons, and the lack of an ideal conduit.⁵

The aim of the present study was to review our 10-year-experience using various esophageal replacement techniques and to compare the complications and outcomes of those procedures in children at the Division of Pediatric Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand.

Ethical approval for this study as a retrospective evaluation of practice was obtained from the Siriraj Institutional Review Board (Si 628/2016).

MATERIALS AND METHODS

The medical records of all patients who underwent an esophageal replacement procedure between 2006 and 2016 were retrospectively reviewed. The clinical data extracted included patient demographics, indications for surgery, perioperative courses, complications and outcomes. Each parameter was reported as a percentage and categorized according to the surgical procedure performed. The patient demographics included the associated congenital anomalies and pre-replacement surgical procedures. The early postoperative outcomes were evaluated according to the duration of the overall admissions, the degree of intensive care and intubation needed, and the number of readmissions after the replacement operation. The perioperative adverse events comprised respiratory complications, esophageal leakages, esophagocutaneous fistulas, gut obstructions, delayed gastric function, dumping syndrome, wound infections and death; they were reported as percentages for each surgical procedure. The operative outcomes were categorized into gastrointestinal and respiratory outcomes and listed in the same fashion. The long-term anthropometric assessments were expressed in degrees of malnutrition, using the weight at the final follow-up compared to the standard growth chart.

RESULTS

Between January 2006 and December 2016, 15 patients underwent an esophageal replacement at the hospital. The patients had mostly been referred from other hospitals. The mean age of the patients was 2.9 years (range: 0.2-15 years). There were 11 (73%) males and 4 (27%) females. The average follow-up duration was 3.6 years (range: 0.4-8 years). The operative procedures of the 15 children studied consisted of 7 (47%) isoperistaltic gastric tubes, 3 (20%) colonic interpositions, 3 (20%) gastric transpositions and 2 (13%) reversed gastric tubes. The indications for esophageal replacement included 5 (33%) long-gap esophageal atresias, 6 (40%) esophageal atresias with postoperative complications, and 4 (27%) caustic injuries. The indications for surgery, categorized by procedure, are demonstrated in [Table 1](#).

The majority of patients (73%) had esophageal atresia, either with long-gap or severe postoperative complications after esophagoesophagostomy (anastomotic leakages, strictures unresponsive to dilatation, empyema thoracis and diverticulum with bleeding), whereas 4 patients (27%) had caustic injuries.

The patient characteristics and associated anomalies are summarized in [Table 2](#). The median age at surgery was 12 months for those patients who underwent the gastric tube and gastric transposition procedures, but much older (108 months) for the colonic interposition group. Accordingly, the median body weights at surgery were 9.6 and 8 kg for the isoperistaltic gastric tube and gastric transposition groups, respectively. The smallest baby in each group weighed 4.7 and 6.2 kg, respectively. There were only 2 cases in the reversed gastric tube group weighing 12 and 21 kg at the time of surgery. As the colonic interposition procedure was performed in older children, their median weight was 20 kg. VACTERL association (vertebral, anorectal, cardiac, renal and limbs anomalies) was the most common associated anomaly, due to esophageal atresia (types A, B and C) being the main indication for surgery.

TABLE 1. Indications for esophageal replacement.

Diagnosis	Isoperistaltic gastric tube	Reversed gastric tube	Gastric transposition	Colonic interposition
Long-gap EA	4	-	-	1
EA with complications	2	1	3	-
Caustic injury	1	1	-	2
Total	7 (47%)	2 (13%)	3 (20%)	3 (20%)

Abbreviation: EA= esophageal atresia

TABLE 2. Patient characteristics.

	Isoperistaltic gastric tube (n = 7)	Reversed gastric tube (n = 2)	Gastric transposition (n = 3)	Colonic interposition (n = 3)
Median age at surgery (months)	12 (3–120)	12, 120	12 (6–24)	108 (36–180)
BW at surgery (kg)	9.6 (4.7–27)	12, 21	8 (6.2–10)	20 (17–23)
Caustic injury	1 (14%)	1 (50%)	-	2 (67%)
Type atresia	6 (86%)	1 (50%)	3 (100%)	1 (33%)
No fistula (type A)	2 (33%)	-	-	1 (100%)
Proximal fistula (type B)	2 (33%)	-	-	-
Distal fistula (type C)	2 (33%)	1 (100%)	3 (100%)	-
Congenital anomaly				
Vertebral	-	-	2 (67%)	1 (33%)
Cardiac	2 (28%)	-	3 (100%)	-
Anorectal	-	-	1 (33%)	-
Renal	-	-	1 (33%)	-
Limbs	-	-	1 (33%)	-
Duodenal atresia	1 (14%)	-	1 (33%)	-
Down syndrome	1 (14%)	-	-	-
Pre-replacement surgery				
Esophagoesophagostomy	2 (28%)	1 (50%)	2 (67%)	-
Gastrostomy	7 (100%)	2 (100%)	3 (100%)	2 (67%)
Cervical esophagostomy	6 (86%)	1 (50%)	3 (100%)	-
Endoscope with dilatation	2 (28%)	1 (50%)	-	2 (67%)
Gastrojejunostomy	-	-	-	1 (33%)
Duodenoduodenostomy	1 (1–4%)	-	-	-

All patients with long-gap esophageal atresia unsuitable for elongation techniques underwent cervical esophagostomy and feeding gastrostomy prior to their replacement surgery. Other types of esophageal atresia patients initially underwent esophagoesophagostomy and subsequently required cervical esophagostomy and gastrostomy due to severe complications, as mentioned above. In the case of children with caustic strictures, 4 (27%) underwent esophageal replacement when their endoscopic dilatations failed. Almost all patients (93%) were fed by gastrostomy until the appropriate time and weight for the replacement procedure.

The early postoperative outcomes are presented in [Table 3](#). The gastric transposition group had the longest median admission period of 68 days owing to previous operations and complications. The intensive care periods after the replacement operation for all groups were typically 2 to 3 weeks. The median intubation period for these techniques was about 2 to 6 days. Postoperative

readmissions occurred approximately 3 to 5 times for esophagoscopy and anastomotic dilatation, although some were due to respiratory problems.

The perioperative complications are summarized in [Table 4](#). There was no perioperative mortality, and none of the grafts were lost. Respiratory complications were the most common adverse event in all groups. There was 1 esophageal leakage (in the gastric transposition group) and 2 esophageal fistulas (one each in the gastric transposition and isoperistaltic gastric tube groups). The complications of delayed gastric function and dumping syndrome arose after gastric transposition, with an incidence of 33% each. Wound infections developed most frequently in the colonic interposition group, affecting all of its cases. All esophageal leakage, esophagocutaneous fistulas and gut obstructions were successfully treated conservatively. The gastrointestinal and respiratory outcomes are listed in [Table 5](#). Full oral feeding was achieved by 86% of the isoperistaltic gastric tube group and 100% of the other

TABLE 3. Early postoperative outcomes.

	Isoperistaltic gastric tube (n = 7)	Reversed gastric tube (n = 2)	Gastric transposition (n = 3)	Colonic interposition (n = 3)
Admission period	37 (17–106)	18, 27	68 (24–84)	22 (20–55)
Intensive care period	17 (8–44)	8, 15	23 (9–35)	13 (4–14)
Intubation period	2 (1–9)	1, 2	6 (1–10)	2 (1–12)
Postoperative readmissions (number of readmissions)	5 (1–14)	2, 12	5 (2–20)	3 (1–3)

Values expressed as days: median (range).

TABLE 4. Perioperative complications.

	Isoperistaltic gastric tube (n = 7)	Reversed gastric tube (n = 2)	Gastric transposition (n = 3)	Colonic interposition (n = 3)
Respiratory complications	1 (14%)	2 (100%)	3 (100%)	1 (33%)
Pneumothorax	-	1 (50%)	-	-
Pneumonia or aspiration	1 (14%)	1 (50%)	3 (100%)	1 (33%)
Esophageal leakage	-	-	1 (33%)	-
Esophagocutaneous fistula	1 (14%)	-	1 (33%)	-
Gut obstruction	2 (28%)	-	-	1 (33%)
Delayed gastric function	-	-	1 (33%)	-
Dumping syndrome	-	-	1 (33%)	-
Wound infection	1 (14%)	-	-	3 (100%)
Death	- (0%)	- (0%)	- (0%)	- (0%)

TABLE 5. Gastrointestinal and respiratory outcomes of esophageal replacement.

	Isoperistaltic gastric tube (n = 7)	Reversed gastric tube (n = 2)	Gastric transposition (n = 3)	Colonic interposition (n = 3)
Gastrointestinal				
Full oral feeding	6 (86%)	2 (100%)	3 (100%)	3 (100%)
Anastomotic stenosis	6 (86%)	1 (50%)	3 (100%)	3 (100%)
Number of endoscopic dilatations per patient	2 (2–11)	6	4 (2–10)	2 (1–31)
Dysphagia	4 (57%)	1 (50%)	1 (33%)	2 (67%)
Oromotor dysfunction	-	1 (50%)	3 (100%)	-
Reflux	1 (14%)	-	1 (33%)	1 (33%)
Tortuous/redundant	2 (28%)*	1 (50%)*	-	3 (100%)
Respiratory				
Chronic lung disease	1 (14%)	-	-	-
Recurrent pneumonia	1 (14%)	1 (50%)	3 (100%)	-
Restrictive lung disease	-	-	-	1 (33%)

*The tortuosity of the grafts was surgically corrected by manubrium excision

groups. Anastomotic stenosis was found in 50%-100% of the cases in the groups, and all resolved after endoscopic dilatation. The median number of endoscopic dilatations required ranged from 2-6 times per patient. Dysphagia and reflux manifested sporadically in 14%-67% of the cases in the groups. Oromotor dysfunction developed in all patients who underwent gastric transposition. Nevertheless, the majority of patients achieved full oral feeding. Tortuosity of the cervical anastomosis arose in 28%-50% of cases, and mainly in the isoperistaltic and reversed gastric tube groups; all cases were surgically corrected by manubrium resection. Redundancy of the graft was present in the colonic interposition group with minimal symptoms; therefore, no intervention was required. The respiratory outcomes comprised chronic lungs, restrictive lung disease and, the most common, recurrent pneumonia (which was found in all patients in the gastric transposition group).

The long-term anthropometric outcomes at the final follow-up are illustrated in [Table 6](#) and [Fig 1](#). The patients had various degrees of malnutrition, particularly in the gastric tube and gastric transposition groups, with 57%-100% of the aforementioned groups weighing less than the third centile on the standard growth chart. An average weight gain was achieved within the study period by 67% of the patients in the colonic interposition group and 20% of the study cohort.

DISCUSSION

Esophageal replacement remains a major challenge in children. Since there is presently no replacement technique that can replace the features of a normal esophagus, many pediatric surgeons believe patients are best served by their own esophagus. The common

indications in children are long-gap esophageal atresia; severe peptic ulcers; caustic and anastomotic strictures; and some rare esophageal disorders such as tumor, prolonged impaction of radiolucent foreign bodies and intractable achalasia.^{5,6,8,9} The common indications for replacement procedure in our study were long-gap esophageal atresia (types A and B) and complicated esophageal atresia with distal tracheoesophageal fistula. Caustic injuries following failed dilatations are another common indication for esophageal replacement in children. As in other developing countries¹⁰, caustic injuries in Thailand continue to be one of the most common health hazards because caustic agents, such as household cleaning materials, are frequently stored in unsuitable or poorly labelled secondary containers.

Various esophageal replacement grafts and techniques have comparable outcomes, none of which emulate the normal esophagus.¹¹ The ideal esophageal conduit should maintain the entire esophageal length, which would allow normal swallowing while technically being simple and adaptable for small children. Accordingly, it should not compress the mediastinum or suppress respiration, should not become tortuous or redundant, should have minimal gastro-esophageal reflux, should not increase the malignancy risk, and should function normally for the lifetime of the patient.^{5,10}

Many esophageal replacement techniques are being practiced and recommended without clear consensus. Furthermore, no randomized, controlled trials have yet demonstrated significant, superior, clinical outcomes of any one of the different types of esophageal replacement. The four most commonly used esophageal replacement techniques are gastric transposition, gastric tube interposition (isoperistaltic or reversed), colonic interposition and

TABLE 6. Long term anthropometric outcomes at final follow up.

	Isoperistaltic gastric tube (n = 7)	Reversed gastric tube (n = 2)	Gastric transposition (n = 3)	Colonic interposition (n = 3)
Malnutrition	4 (57%)	2 (100%)	2 (67%)	1 (33%)
Percentile body weight				
< 3 percentile	4 (57%)	2 (100%)	2 (67%)	1 (33%)
3-25 percentile	2 (28%)	-	1 (33%)	-
25-75 percentile	1 (14%)	-	-	2 (67%)
75-97 percentile	-	-	-	-

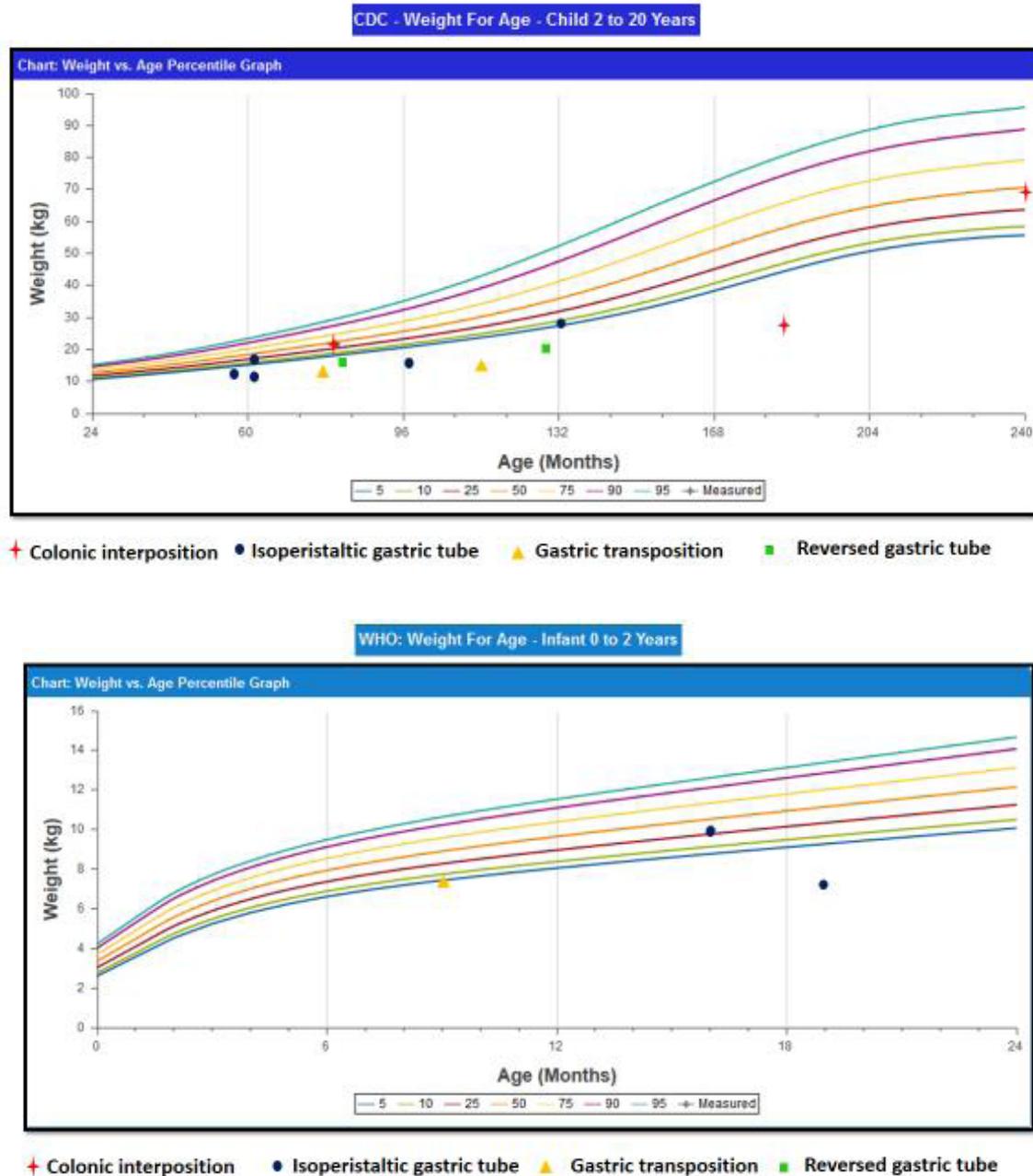


Fig 1. Growth at final follow-up.

jejunal interposition.^{8,11-15} At our institute, the more technically demanding jejunal interposition has been the least preferred; therefore, the esophageal replacement procedures practiced have comprised gastric transposition, isoperistaltic or reversed gastric tube, and colonic interposition. Almost half (47%) of the patients studied at our institute underwent isoperistaltic gastric tube, given that the stomach has better vascularity and fewer ischemic complications.^{1,11} The graft choices were based on anatomical considerations and the preferred technique of the treating surgeons rather than on any discernible objective data. The factors influencing the outcomes were

related to the relatively infrequent need for esophageal replacement, the surgeons' expertise and the absence of ideal grafts.⁵

Various esophageal substitutes have different advantages, technical difficulties and specific complications. The overall morbidity is high, the most common being anastomotic leakage and stricture, with both ranging from 10% to 20%.⁸

The overall replacement-related mortality rate has been assessed to be approximately 2%.⁵ In our study, there was no perioperative mortality, and none of the grafts were lost. The early post-operative outcomes of the

methods showed no differences in their intubation periods, intensive care periods or post-operative readmission rates. The intubation period was about 1-2 days, whereas the intensive care period was typically about two weeks. Patients in the gastric transposition group had the longest median intubation and intensive care periods (6 and 23 days, respectively), being complicated by type-C esophageal atresia from previous esophagoesophagostomy operations with associated cardiac anomalies. The post-operative readmissions of all groups were due to pneumonia or esophageal dilatation, and they averaged around 3-5 times per group.

Gastric tube interposition grafts are constructed from the greater curvature in a reversed (antiperistaltic) or isoperistaltic fashion.⁹ Tube graft necrosis is rare because the gastric tube has an excellent and reliable blood supply from the submucosal plexus and gastroepiploic vessels.¹ The gastric tube can bridge relatively long gaps and remain as a passive conduit with a tubular shape and without dilatation or tortuosity. Other series using a gastric tube substitute had cervical anastomotic leakage despite being sealed spontaneously with dilatable strictures.^{1,16} In our study, the most common perioperative adverse event was respiratory complications, appearing in both isoperistaltic and reversed gastric tube interpositions. Esophagocutaneous fistulas (14%), gut obstructions (28%) and wound infections (14%) were not common after isoperistaltic gastric tube interposition, and all were resolved by conservative treatment.

The evaluation of the long-term gastrointestinal outcomes found that 86% of the patients in the isoperistaltic gastric tube interposition group and all patients in the other groups achieved full oral feeding. Anastomotic stenosis was common, albeit dilatable in all groups. Borgnon et al.¹ reported that of their series of esophageal replacements with isoperistaltic gastric tube, 80% achieved a normal diet, 15% had mild dysphagia, 15% had major dysphagia, and 10% had redundant grafts with dumping syndrome and cervical Barrett's esophagus. Our study demonstrated that 57% of the patients had dysphagia, 14% had reflux and 28% had tortuosity of the cervical anastomosis. In consideration of the proximal esophageal graft anastomosis, most could be achieved through the neck incision. When the anastomosis is high within the neck, the thoracic inlet can act as a constriction to the anastomosis. In that event, the thoracic inlet can be widened by resecting the upper part of the manubrium or the sternal head of the left clavicle. Consequently, the tortuous grafts in 3 patients who underwent gastric tube interposition procedures were surgically corrected

by manubrium excision. Although the incidences of long-term respiratory problems, including recurrent pneumonia and chronic lung diseases, ranged from 14%-50%, they did not differ between the two gastric tube groups.

The disadvantage of the gastric tube is an extensive suture line that produces a higher incidence of leakages and strictures. The reduced gastric capacity and the production of acid within the tube graft results in acid reflux into the cervical esophagus, precipitating Barrett's esophagitis.^{1,5,16,17} Esophagitis and metaplasia have been described in children following gastric tube replacement. There should be regular monitoring of the esophageal conduit since chronic exposure to acid reflux may predispose to metaplasia and adenocarcinoma.¹

Gastric transposition or the gastric pull-up procedure has several advantages. It is relatively simple with a single anastomosis at the neck, creating a sufficiently long and very well-vascularized graft.^{10,12} It has been shown to have a reduced risk of anastomotic leakage, stricture and necrosis.^{5,8} The perioperative complications in our study were esophageal leakage, esophagocutaneous fistula, delayed gastric function and dumping syndrome (each arising in 33% of patients) and pneumonia (in all patients). Vagotomy during gastric mobilization may lead to delayed gastric emptying and dumping syndrome. In addition, pyloromyotomy and pyloroplasty may be performed to prevent delayed gastric emptying. Spitz et al.^{8,18} reported the outcomes of gastric transposition in 236 patients, which were a 2.5% mortality rate, a 12% leakage rate and a 20% stricture rate. In our study, the long-term outcomes of the gastric transposition group revealed all patients (100%) achieved full oral feeding, with all having anastomotic stenosis, oromotor dysfunction and recurrent pneumonia. It is speculated that the recurrent pneumonia was a consequence of underlying complicated, type C, esophageal atresia. However, previous studies have acknowledged that gastric transposition is associated with a higher respiratory morbidity.¹¹ Occupying the mediastinum and chest, the stomach may not empty effectively, causing compression (mass effect) of the intrathoracic organs and a long-term reduction of the lung capacity. The long-term gastrointestinal outcomes also included 33% gastroesophageal reflux and dysphagia rates. The reflux may lead to recurrent heart burn, regurgitation, bad breath and pneumonia. Dysphagia has been found to be common, despite successful and adequate replacements^{5,6}, which could be due to discoordinated peristalsis, antiperistaltic layout, a tortuous esophageal conduit or significant acid reflux.

Colonic interposition is the most frequently used esophageal replacement procedure and provides superior length.^{1,5} The perioperative complications in our study showed 33% respiratory complication and gut obstruction rates. Although there was a 100% wound infection rate, there were no leakages or fistulas. In the long-term follow-up, all patients (100%) achieved full oral feeding, and they developed dilatable anastomotic stenosis and redundancy of the colonic graft; however, the patients did not require intervention. The long-term gastrointestinal outcomes comprised dysphagia (67%) and reflux (33%), while the respiratory problem was restrictive lung disease (33%). In 2015, Lobeck et al.¹⁴ reported that the most common postoperative complication among 10 patients after colonic interposition was esophageal stricture (54%). The disadvantage of colonic interposition is the redundancy of grafts with stasis and dysphagia due to a negative pressure in the thoracic cavity and emptying by gravity.^{4,5,19, 20} Complications such as anastomotic leakage and strictures have also been reported to be related to a precarious blood supply.⁴ Other renowned, large-scale series^{8,15,18} with long-term outcomes have demonstrated no significant differences in the early or late complications of the gastric transposition and colonic interposition techniques.

Jejunal interposition can be used as a pedicle or free graft.¹¹ The main advantage of this graft type is the most suitable caliber of the grafts with peristaltic activity.³ Conversely, high failure, morbidity and mortality rates¹³ have been reported for this procedure, presumably due to technical difficulties and a tenuous blood supply. Our center has no experience with this technique.

The average follow-up duration in the current study was 3.6 years (range: 0.4-8 years). The measured growth in patients after esophageal replacement at the final follow-up demonstrated growth retardation and malnutrition in all procedural groups, with 57% in the isoperistaltic gastric tube, 100% in the reversed gastric tube, 67% in the gastric transposition and 33% in the colonic interposition groups. Malnutrition was deemed to be present when a child's weight-for-age fell below the third centile of the standard growth chart. Growth retardation is prevalent in patients with esophageal atresia. Oral aversion is common among infants with long-gap esophageal atresia due to delayed oral feeding, caused by either a lack of sham feeding in oral feeds or delays in the replacement.⁵ It is important to monitor nutritional status, growth and development in children after an esophageal replacement as they are known to fall below centiles for both height and weight.^{1,3,4}

The limitations of a retrospective design, the small number of patients and technical variations led to difficulty in making comparisons to discover any statistical significance in this study. Close monitoring and long-term follow-up through more substantial group studies may elicit the clinically important factors relating to the care of these patients.

CONCLUSION

Esophageal substitution remains a major challenge in children. Our experience indicates that children can be safely operated on using any of the various methods currently available, with acceptable perioperative morbidities and no mortality. Nevertheless, the long-term consequences and complications should be monitored throughout adulthood.

Conflict of interest: The authors have no conflicts of interest to declare.

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