

Outcomes and Satisfaction of Esophageal Replacement in Children at Queen Sirikit National Institute of Child Health: A 10-year Review

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Abstract

Objectives: Esophageal replacement (ER) is a major operation and not frequently preformed especially in children. The aim of this study is to review the outcomes of patients who underwent ER in a 10-year period and evaluate satisfactions of patients or parents after the procedures.

Materials and Methods: Retrospective chart review of all patients who underwent ER at Queen Sirikit National Institute of Child Health during 2007-2016 was conducted. Demographics, causes of ER, types of ER, postoperative outcomes and satisfaction results were collected.

Results: A total of 22 patients underwent ER during the study period. The most common indication for ER was isolated esophageal atresia 13 cases (59.1%) followed by caustic esophageal stricture in 6 case (27.2%). Mean age at the operation was 28 months. Gastric transposition (GT) was the most common procedure performed in 15 cases, followed by colonic interposition (CI), and Reverse gastric tube esophagoplasty (RGT). Over half of cases (59.1%) underwent ER via posterior mediastinal route especially in GT and RGT. Common immediate postoperative complications were pneumonia, wound infection and anastomotic leakage. Mean postoperative initial feeding was 11 days (range 10-14 days). Mean hospital stay was 42 days (range 12-105 days). There was no mortality in all of the patients. For the long-time follow-up, 90% of the patients had appropriate development of age with ability to eat normally and satisfactory quality of life.

Conclusion: The outcomes of ER in children at Queen Sirikit National Institute of Child Health institute is excellent. There are minimal post-operative complications without no mortality. There were no statistic difference of postoperative complications and patient's development amongst various ER procedures. Long-term post-operative course is satisfactory.

Keywords: Esophageal replacement, Gastric transposition, Reverse gastric tube esophagoplasty, Outcome, Satisfaction

INTRODUCTION

Esophageal replacement (ER) is a major operation but rarely performed especially in children. There are many procedures for ER, in each procedure there are ad-

vantages and disadvantages since up to date there is not a single optimum standard procedure^{1,2}. Children whom had undergone ER are still expected to have a long-life expectancy with the best quality of life possible³.

OBJECTIVES

The aim of this study is to review the outcomes of patients whom underwent esophageal replacement in a 10-year period. Focusing on post-operative complications, long term post-operative course, functional outcome and patient's and/or parent's satisfactions after the procedure.

MATERIALS AND METHODS

We reviewed the medical records of all patients whom underwent ER at Queen Sirikit National Institute of Child Health (QSNICH), a tertiary pediatric center, from January 2007 to December 2016. Inclusion criteria was all patients under 15 years of age whom underwent ER at our institute, excluding all patients whom received prior surgeries from other hospitals. The surgery was performed by 7 experienced pediatric surgeons.

Data collection was done by separating the data into 2 periods; immediate period and long-term period. Immediate period was the data collected during the in-hospital care for ER which were demographic data, gender, age at the time of ER, associated anomalies, indication for ER, type of ER, route of surgery, complications, length of hospital stay, post-operative initial feeding day, mode of post-operative feeding, and mortality.

Long-term period was the data collected during the follow-up post-ER which were growth and development, functional outcome, medication requirement and patient's and/or parent's satisfactions after the procedure to state the patient's quality of life. For the evaluation

of the nutritional status; Waterlow classification⁴ was used. As for the patient's development assessment, it was evaluated by a pediatrician during routine follow-up.

For satisfaction evaluation, a numeric scale questionnaire was used to assess satisfaction after surgery. It was obtained in person at the time of the latest follow-up or via telephone using a single interviewer. If these two methods were not possible then a postal letter was sent to the patient's home. The numeric scale questionnaire used was a dropdown numeric 1-5 scale which 1 is a terrible result and 5 is an excellent result. Patients whom were 11 years and older or caregivers are asked to answer the questionnaire. There were 7 questions. The questions asked were focused at the satisfaction of the patient's growth and development, the ability to eat normally, overall quality of life, the cosmetic appearance of the surgical scar and chest deformities, and regarding the long-term medical care including follow-up time and the medical provider.

Data analysis was done using mean and percentage. Then data comparison was done using Pearson Chi-square and Fisher's exact test. Statistical significance was defined as *p*-value less than 0.05. This is a retrospective descriptive study. The study was approved by the Research Ethics Committee of our institute.

RESULTS

Twenty-two patients underwent esophageal replacement in a 10-year period (Table 1).

Table 1 Demographic data

Patient characteristics (N=22)	GT (n=15)	RGT (n=3)	CI (n=4)	<i>p</i> -value
Gender				
- Male (%)	9 (60)	1 (33.3)	4 (100)	0.603
- Female (%)	6 (40)	2 (66.7)	0	
Mean age at the time of ER (months)	15	23	83	0.002
Associated anomalies				
- VACTERL association (%)	2 (13.3)	0	0	0.619
- Cardiac anomalies (%)	1 (6.7)	0	0	
- Tracheomalacia (%)	3 (20)	0	0	
Indication for ER				
- Isolated esophageal atresia (%)	11 (73.3)	2 (66.7)	0	
- Long gap esophageal atresia (%)	2 (13.3)	0	0	
- Caustic esophageal stricture (%)	2 (13.3)	1 (33.3)	3 (75)	
- Esophageal duplication (%)	0	0	1 (25)	

ER: Esophageal Replacement; GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition

There was male predominance of 1.75 to 1. The most common indication for ER was isolated esophageal atresia (59%) followed by caustic esophageal stricture (27.2%). Mean age of patients at the time of ER was 28 months (range 3 months to 11.9 years). The patients whom underwent colonic interposition (CI) were significantly older than patients whom underwent gastric transposition (GT) and reversed gastric tube esophagoplasty (RGT). Patients whom received CI were indicated by caustic esophageal stricture and esophageal duplication. The most common associated anomalies were tracheomalacia ($n = 3/22$, 13.6%), VACTERL association ($n = 2/22$, 9%) and cardiac anomalies ($n = 1/22$, 4.5%), respectively.

There were 3 procedures used for ER: gastric transposition (GT), reversed gastric tube esophagoplasty (RGT) and colonic interposition (CI) (Table 2). Stomach (81.8%) was the most common graft used to replace the esophagus which GT was the most common procedure performed. There were three routes used for these ER:

retrosternal, transpleural and posterior mediastinal route. The most common route for GT and RGT procedure was via posterior mediastinal route. As for CI procedure, the retrosternal route was most commonly used.

Immediate outcomes of ER (Table 3) were favorable, there were no deaths with minimal complications. The most common complication was pneumonia ($n = 12/22$, 54.5%), wound infection ($n = 5/22$, 22.7%) and anastomotic leakage ($n = 3/22$, 13.6%), respectively. For the 3 patients whom developed anastomotic leakage, all 2 patients of GT were managed successfully by conservative management. However, the patient of CI failed conservative management and required a reoperation. One patient of GT had pneumothorax which required an intercostal drainage tube insertion. Another patient of GT had intestinal obstruction due to adhesion which also required a reoperation. There was neither no graft necrosis nor laryngeal nerve injury. Regarding enteral nutrition, most of all patients whom underwent ER were discharge home with full oral feeding (81.8%), despite

Table 2 Type of esophageal replacement procedures and route of surgery

Route of Surgery	Gastric graft (n=18)		Colonic graft/CI (n=4)
	GT (n=15)	RGT (n=3)	
Retrosternal (%)	0	0	2 (50)
Transpleural (%)	6 (40)	0	1 (25)
Posterior mediastinal (%)	9 (60)	3 (100)	1 (25)

GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition

Table 3 Immediate outcomes of esophageal replacement

N=22	GT (n=15)	RGT (n=3)	CI (n=4)	p-value
Complication				
- Pneumonia (%)	6 (40)	3 (100)	3 (75)	0.195
- Wound infection (%)	3 (20)	0	2 (50)	0.316
- Anastomotic leakage (%)	2 (13.3)	0	1 (25)	0.705
- Intestinal obstruction (%)	1 (6.7)	0	0	0.738
- Pneumothorax	1 (6.7)	0	0	0.738
Mean hospital stay (day)	44	24	47	0.338
Mean post-operative initial feeding day	10	12	14	0.115
Mode of post-operative feeding				
- Oral (%)	13 (86.7)	2 (66.7)	3 (75)	0.545
- Gastrostomy tube (%)	0	1 (33.3)	1 (25)	0.023
- Jejunostomy tube (%)	1 (6.7)	0	0	0.738
- Nasojejunum tube (%)	1 (6.7)	0	0	0.738

GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition

the type of ER. Only 4 patients were discharge home with tube feeding: 2 via gastrostomy, 1 via jejunostomy and 1 via nasojeunal tube. The mean time of initiating enteral feeds were 11 days (range 8-25 days). Mean hospital stay was 42 days (range 12 - 105 days). There was no significant difference between the types of ER and complications, post-operative initial feeding day and hospital stay.

Long-term outcomes of ER (Table 4), there was no lost to follow-up. Mean follow-up time was 3.7 years (range 2 months - 10.3 years). Sixty-four percent of all patients had malnutrition grade 1 according to

Waterlow's Classification (Figure 1). However, 90% of all patients had appropriate development to age. There was no statistical difference between the type of ER and nutritional status or development. Regarding functional outcomes, 18% of all patients had respiratory, and/or cardiac function impairment. One (4.5%) patient whom had VACTERL association with the underlying cardiac anomaly had respiratory and cardiac function impairment. The other 3 (13.6%) patients had respiratory function impairment with multiple admissions due to pneumonia. For gastrointestinal (GI) functional impairment, 5 (22.7%) patients had dysmotility, 4 (18%)

Table 4 Long-term outcomes of esophageal replacement

N=22	GT (n=15)	RGT (n=3)	CI (n=4)	p-value
	(%)	(%)	(%)	
Development assessment				
Global delayed	2 (13.3)	0	0	-
Respiratory and cardiac function				
Impaired	2 (13.3)	0	2 (66.7)	0.139
Gastrointestinal function				
Gastroesophageal reflux	2 (13.3)	1 (33.3)	0	0.279
Dysphagia	2 (13.3)	0	0	0.738
Dysmotility	2 (13.3)	1 (33.3)	2 (50)	0.212
Constipation	2 (13.3)	2 (66.7)	0	0.598
Anastomotic stricture	0	1 (33.3)	1 (25)	0.091
Mode of enteral feeding				
- Oral	15(100)	2 (66.7)	3(75)	0.091
- Gastrostomy	0	1 (33.3)	1(25)	0.286
Medication				
- Antacid	0	1 (33.3)	1 (25)	0.091
- Proton pump inhibitor	4 (26.7)	1 (33.3)	1 (25)	0.966
- Prokinetic drug	3 (20)	0	1 (25)	0.662

GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition

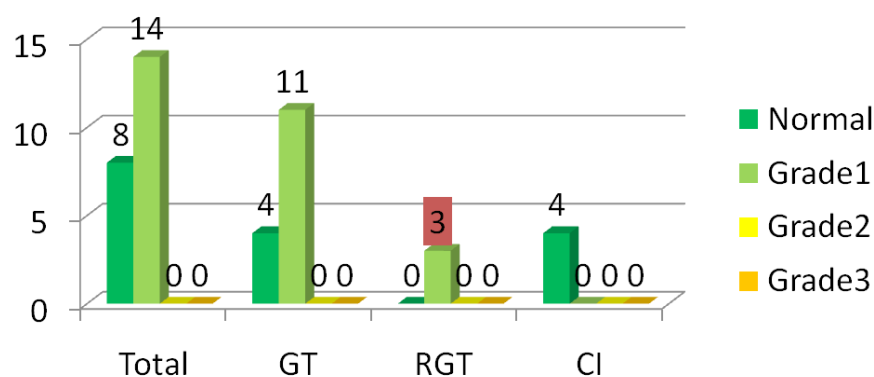


Figure 1 Waterlow classification (%Weight/Height)

patients had constipation and 3 (13.6%) patients had gastroesophageal reflux (GERD). Half of all patients were on medications for GI function impairment which proton pump inhibitor (PPI) drugs were most commonly prescribed. Anastomotic stricture developed in 2 patients (9%) which responded to endoscopic dilatation, one patient each underwent RGT and CI. Despite of these GI function impairment, 90% of all patients had normal oral feeding. There was no statistical significance between type of ER and functional impairment or enteral feeding or home medications.

Satisfaction assessment after via questionnaire was obtained (Table 5). Despite there were no lost to follow-ups, however many families were not reachable during the survey period, only 63% answered the questionnaire. However, the results were great. The patients and caregivers were satisfied in all aspects: patient's growth and development, ability to eat normally, overall quality of life, cosmetic appearance of the surgical scar and chest deformities, and long-term medical care including follow-up time and medical provider.

Table 5 Satisfaction assessment

N=14 (63%)	Percentage of satisfaction (%)
Growth and development	81
Eating	87
Quality of life	88
Chest contour	91
Surgical scar	94
Follow-time	94
Surgeon	100

DISCUSSION

The outcomes of esophageal replacement during this 10-year period was excellent. There was no mortality (Table 6). This was possible due to the improvement of specialized neonatal and pediatric care especially optimal intensive care, parenteral nutrition and the meticulous surgical technique despite the type of ER and modernized surgical equipment. However, these data might be influenced by the different indications for ER; caustic stricture compared to esophageal atresia or different age group; younger in patients with esophageal atresia and older in patients with caustic esophageal stricture or surgeon's preference to choose the type of ER procedure. The optimal method of esophageal replacement still remains controversial^{1,2,5}.

Overall, the posterior mediastinal route was the preferred approach as in other series^{5,6}, despite the type of ER performed because of the shortest distance and no compression effect to the lungs. However, pneumothorax is the complication caused by blunt dissection of the mediastinal bed, our study occurred 1 case of GT (6.7%) much lower than the 17.6% of the GT group from Tannuri et al study⁷. This is a minor immediate complication treated by intercostal drainage. For major immediate complications post ER, we did not have graft necrosis and laryngeal nerve injury during this 10-year period. Anastomotic leakage was the third common immediate complication post ER. From our study, 13.6% of all patients had anastomotic leakage which is quite comparable to the 12% of Spitz et al^{5,6} in 2009 and 2014. It is also found that the leakage is more frequent in colon conduit^{7,8}. However, pneumonia (54.5%) was our most common immediate complication despite the indication for ER, this is the most concerning post-operative issue to be dealt with since all patients required ventilator support. Burgos et al⁸ found only 10.4% from all post ER patients that had pneumonia.

Table 6 Comparison of mortalities and complications post esophageal replacement

Study	N	Type of ER	Mortality (%)	Graft necrosis (%)	Anastomotic leakage (%)
Tannuri ⁷ , 2007	149	GT, CI	2	1.3	26.1
Spitz ⁶ , 2009	192	GT	4.6	0	12
Burgos ⁸ , 2010	96	CI	9.3	2	23.9
Spitz ⁵ , 2014	236	GT	2.5	0	12
This study, 2018	22	GT, RGT, CI	0	0	13.6

ER: Esophageal Replacement; GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition

Table 7 Comparison of gastrointestinal functional impairment post esophageal replacement

Study	N	Type of ER	Dysmotility (%)	GERD (%)	Dysphagia (%)	Esophageal stricture (%)
Tannuri ⁷ , 2007	149	GT, CI	0	15.6	0	14.7
Coopman ⁹ , 2008	32	CI	N/A	20	50	28
Spitz ⁶ , 2009	192	GT	8.7	N/A	N/A	19.6
Burgos ⁸ , 2010	96	CI	N/A	43	N/A	11.4
Spitz ⁵ , 2014	236	GT	8.8	N/A	N/A	20
Lima ¹⁰ , 2015	72	CI	N/A	70.8	25	6.9
Koivusalo ¹¹ , 2017	11	RGT	N/A	80	38	18
This study, 2018	22	GT, RGT, CI	22.7	13.7	9	9

ER: Esophageal Replacement; GT: Gastric Transposition; RGT: Reversed Gastric Tube; CI: Colonic Interposition; GERD: Gastroesophageal Reflux Disease

Focusing on the outcome post esophageal replacement, we noted that there was no direct correlation between immediate and long-term outcomes. Patients whom developed complications soon after ER do not necessarily have problems seen on long-term follow-up.

According to literature, the main long-term complications of ER are GI functional impairment. Coopman et al⁹ reported 18 patients (56.2%) in all 32 patients post CI had digestive symptoms. Our mean follow-up time was 3.7 years. In this period, our patients had higher dysmotility than other series (table 7). However, GERD, dysphagia, esophageal stricture was much lower. The dysmotility, GERD and dysphagia were all managed by medication, no additional antireflux procedures. The patient's symptoms usually relieved during long term follow-up. Therefore, the role of antireflux surgery may be clarified in future^{10,11}. There was no statistical significance between the type of ER and these GI functional impairments. Regarding esophageal stricture which were quite lower than other series, it was all managed by dilatations, no reoperation required. It was noted that developing post ER esophageal stricture was found in patients with caustic injury as the indication for ER which was similar to other studies^{2,5,6}. However, no statistical significance was found.

Nutritional status and quality of life are the concerning issues for long-term outcomes post ER. Being able to grow and have a healthy satisfying quality of life, regardless of the type of ER, is the important goal for both medical providers and parents. Our study found that most of our post ER patients had malnutrition grade 1 (64%) according to Waterlow's Classification. This is because of most patients (70%) were born with

esophageal atresia and low birth weight; less than 2,200 grams, which is also noted by Coran et al^{1,2} that children who originally born with esophageal atresia tend to be in lower percentiles for height and weight. Our patients (87%) were found to have appropriate development to age except patients with significant associated anomalies which are reasonable. The quality of life is satisfactory (88%) which is similar to other series^{3,12,13}. They can go to school and eat trans orally. Also, they are happy about the cosmetics and obviously the medical team.

CONCLUSION

The outcomes post-esophageal replacement in children at our institute is excellent. There are minimal post-operative complications and no deaths. Long term post-operative course is satisfactory. The choice of surgical procedure for esophageal replacement is still by surgeon's preference not by evidence based. Limitation of this study is due to the small sample size and its retrospective nature. Multi-institutional prospective study should be conducted.

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บทคัดย่อ ผลการรักษาและความพึงพอใจ ของการผ่าตัดเปลี่ยนหลอดอาหารในเด็ก ณ สถาบันสุขภาพเด็ก
แห่งชาติมหิดลราชินี ในช่วงระยะเวลา 10 ปี

กมลทิพย์ ยุทธพงศ์, พบ., วรณนิสา ภูเจริญ, พบ., รัชสรรค์ นิรามิช, พบ.

กลุ่มงานศัลยศาสตร์ สถาบันสุขภาพเด็กแห่งชาติมหิดลราชินี กรุงเทพฯ

วัตถุประสงค์: การผ่าตัดเปลี่ยนหลอดอาหารเป็นการผ่าตัดที่สำคัญและมีการทำไม่บ่อยนัก โดยเฉพาะ
ในเด็ก จุดมุ่งหมายของการศึกษาค้นคว้าครั้งนี้เพื่อศึกษาผลลัพธ์ของผู้ป่วยที่ผ่าตัดเปลี่ยนหลอดอาหารในช่วงระยะเวลา
10 ปี และประเมินความพึงพอใจของผู้ป่วย หรือผู้ปกครองภายหลังการทำผ่าตัดไปแล้ว

วัสดุและวิธีการ: เป็นการศึกษาย้อนหลังจากเวชระเบียนของผู้ป่วยที่ได้รับการผ่าตัดเปลี่ยนหลอดอาหาร
ที่สถาบันสุขภาพเด็กแห่งชาติมหิดลราชินี ตั้งแต่ปี พ.ศ. 2550 ถึงปี พ.ศ. 2559 ข้อมูลทั่วไป สาเหตุที่ทำให้ต้องผ่าตัด
เปลี่ยนหลอดอาหาร ชนิดของการผ่าตัดเปลี่ยนหลอดอาหาร ผลของการรักษา และผลการประเมินความพึงพอใจ
ได้ถูกรวบรวมเพื่อการศึกษา

ผลการศึกษา: ผู้ป่วยทั้งหมด 22 ราย ได้รับการผ่าตัดเปลี่ยนหลอดอาหารในช่วงระยะเวลาที่ทำการศึกษา
ข้อบ่งชี้ที่ต้องทำผ่าตัดเปลี่ยนหลอดอาหารที่พบบ่อยที่สุดคือ หลอดอาหารตันแต่กำเนิดชนิดไม่มีรูต่อกับหลอดลม
พบ 15 ราย รองลงมาได้แก่ การตีบของหลอดอาหารจากการกลืนสารกัดกร่อน การผ่าตัดนำกระเพาะอาหาร
ทั้งหมดขึ้นไปในช่องอกเพื่อต่อกับหลอดอาหาร เป็นวิธีการทำผ่าตัดเปลี่ยนหลอดอาหารที่ทำมากที่สุดคือ 15 ราย
รองลงมาคือ การใช้ลำไส้ใหญ่ขึ้นไปต่อระหว่างหลอดอาหารกับกระเพาะอาหาร และการตัดบางส่วนของกระเพาะ
อาหารเย็บเป็นหลอดอาหารขึ้นไปต่อ มากกว่าครึ่งหนึ่งของผู้ป่วยได้รับการผ่าตัดเปลี่ยนหลอดอาหารทางด้าน
หลังของช่องอก โดยเฉพาะการใช้กระเพาะอาหารและบางส่วนของกระเพาะอาหารขึ้นไปต่อกับหลอดอาหาร
ด้านบน การผ่าตัดโดยใช้ลำไส้ใหญ่ขึ้นไปต่อมักจะทำผ่านทางใต้กระดูกหน้าอกทางด้านหน้า ภาวะแทรกซ้อน
หลังผ่าตัดที่พบบ่อยคือ ปอดบวม การติดเชื้อของแผลผ่าตัด และการแยกของรอยเย็บต่อ ไม่มีความแตกต่างกัน
ทางสถิติของภาวะแทรกซ้อนหลังการผ่าตัดระหว่างวิธีการผ่าตัดเปลี่ยนหลอดอาหารทั้งสามแบบ ระยะเวลา
ที่ผู้ป่วยสามารถรับอาหารได้เฉลี่ย 11 วัน (พิสัย 10-14 วัน) ระยะเวลาที่นอนในโรงพยาบาลเฉลี่ย 42 วัน (พิสัย
12-105 วัน) ไม่มีผู้ป่วยเสียชีวิตในการผ่าตัดเปลี่ยนหลอดอาหาร การติดตามผลการรักษาระยะยาวพบว่า
ร้อยละ 90 ของผู้ป่วยมีการพัฒนาการเป็นไปตามอายุของผู้ป่วย พร้อมกับการรับประทานอาหารได้เป็นปกติ
และคุณภาพชีวิตอยู่ในเกณฑ์ที่น่าพอใจ

สรุปผลการศึกษา: ผลการรักษาการเปลี่ยนหลอดอาหารในเด็กที่สถาบันของเราได้ผลดีมาก ไม่มีผู้ป่วย
เสียชีวิต ไม่มีความแตกต่างทางสถิติของภาวะแทรกซ้อน รวมถึงการพัฒนาการของผู้ป่วยหลังการผ่าตัดเปลี่ยน
หลอดอาหารในวิธีต่างๆ และการติดตามผลการผ่าตัดระยะยาวอยู่ในเกณฑ์ที่น่าพอใจ