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ORIGINAL ARTICLE

Therapeutic strategy for thoracoscopic repair of esophageal atresia and its outcome

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Abstract

Purpose Thoracoscopic repair can be safely performed in most types of congenital esophageal atresia (EA), including in patients with long gap EA or very low birth weight. Accordingly, we performed single- or multistage thoracoscopic repair for various EA types. We aimed to report our therapeutic strategy for thoracoscopic radical surgery for treating EA and its outcome.

Methods Outcomes of radical surgeries for treating congenital EA at our institute from 2013 to 2018 were retrospectively evaluated.

Results Thirty-eight radical surgeries were evaluated: 3 Gross type-A, 1 type-B, 30 type-C, 1 type-D, and 3 type-E. The cervical approach was performed in 5 cases and thoracoscopic esophageal anastomosis in 33, including 26 single-stage (all type-C) and 7 multistage surgeries (3 type-A, 3 type-C, and 1 type-D). There were no cases of thoracotomies or intraoperative thoracoscopic surgery complications. Three cases of minor leakage were conservatively resolved. Three postoperative chylothorax surgeries (9%) and seven balloon dilatations (21%) for anastomotic stenosis were performed.

Conclusion Thoracoscopic radical surgery for treating EA, including single- and multistage procedures, can be performed, except in type-E cases or when the end of the proximal esophagus is located higher than the clavicle.

Keywords Congenital esophageal atresia · Thoracoscopic repair · Primary anastomosis · Multistage anastomosis

Introduction

Thoracoscopic repair of esophageal atresia (EA) has been rapidly gaining popularity as it enables a good visual field during surgery and has excellent cosmetic outcomes. Indications for this procedure are broad; however, factors that can influence the procedure, such as body weight and gap length, are still being assessed.

We believe that thoracoscopic repair can be safely performed in most types of EA, including in patients with long gap EA or very low birth weight. Accordingly, we performed single- or multistage thoracoscopic repair for various types of EA. Herein, we describe our treatment strategy for thoracoscopic repair and our experiences with postoperative complications, such as gastroesophageal reflux and congenital esophageal stenosis.

Material and methods

Before study commencement, all protocols were approved by the ethics review board of our institute (Approval Number: 2014-0400). Patients who underwent surgery for EA at our institution from August 1, 2013, to December 31, 2018, were included in the study. Details of the operative process were summarized, and patients were divided into either single-stage or multistage anastomosis groups. Radical surgeries performed included thoracoscopic repair for EA. Esophageal anastomosis under thoracoscopic guidance was performed even for patients with long gap EA, extremely low birth weight (ELBW), and low birth weight (LBW) (1590 g) using the multistage approach. Surgery outcomes were retrospectively evaluated on the basis of observation items including EA classification, concurrent malformation,

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age at the time of surgery, body weight, operative duration, estimated blood loss, intraoperative complications, length of hospital stay, postoperative complications, anastomotic stricture, congenital esophageal stenosis, and chylothorax.

Radical thoracoscopic repair of esophageal atresia

On confirming the disease type before surgery by contrast-enhanced radiography using isotonic iodixanol (VISIPAQUE injection), the site of the tracheoesophageal fistula (TEF) was identified using a fiber-optic bronchoscope following intubation. Surgery was performed with positive-pressure pneumothorax of 4–8 mmHg using two 3-mm ports and one 5-mm camera port. Intubation was generally performed immediately before surgery, and muscle relaxants were not used until ligation of the TEF. In our anastomosis procedure, the first two sutures were started from the mucosa of the distal esophagus using a reverse needle to ensure that the mucous was sutured. We also made sure that the seams were not too fine, and we stitched around the entire circumference of the esophagus using approximately six needles. We used 5–0 polydioxanone RB-3 (PDS; Ethicon Inc., Somerville, NJ) suture for the anastomosis.

Elongation

Thoracoscopic internal esophageal traction was performed during the initial surgery. We applied traction sutures to both ends of the esophagus using 5–0 absorbable sutures and tied each suture around the most suitable costal bone to approximate the esophageal ends [1]. After 1–2 weeks, we performed thoracoscopic delayed esophago-esophageal anastomosis. The period between elongation and delayed primary anastomosis was decided based on previous reports [2–4].

Assessment of esophageal anastomotic structure

Patients were diagnosed with anastomotic stricture under contrast-enhanced radiography if they had a stricture index of > 50% as reported by Said et al. [5]. Balloon dilatation was performed in patients exhibiting any symptoms indicative of anastomotic stricture such as stuck food, regardless of radiographic findings.

Results

During single-stage anastomosis in a patient weighing less than 2000 g, one skilled surgeon is involved. Otherwise, nine pediatric surgeons with anastomosis skills above a certain level in the dry lab are assigned to the operation. There is no other special operator required. A total of 39 patients were treated for EA at our institute during the study period. Radical surgery was performed in 38 cases (Gross type-A: 3 cases, type-B: 1 case, type-C: 30 cases, type-D: 1 case, and type-E: 3 cases), but the remaining patient (type-C) who underwent abdominal esophageal banding and gastrostomy was transferred to another hospital due to concomitant severe cardiac malformation.

Gross type

Gross type-A case

In 2 of 3 cases, thoracoscopic esophageal elongation for a long gap (four or more vertebrae) was performed at the first surgery. In the remaining patient with anorectal malformation, gastrostomy without esophageal elongation and colostomy were performed. This was followed by thoracoscopic esophageal anastomosis at ages of 11, 13, and 34 days (Table 1).

Gross type-B case

In the Gross type-B case with anorectal malformation and vertebrae, anus, trachea, esophagus, and renal associations, esophageal elongation was performed at the first surgery. Repeat thoracoscopic esophageal elongation caused mediastinitis during the second surgery; therefore, we performed laparoscopic-assisted gastric tube reconstruction with cervical esophagogastric anastomosis as the third surgery (Table 1).

Gross type-C cases

Among the 30 Gross type-C cases, thoracoscopic esophageal anastomosis was performed as single-stage and multistage surgery in 26 and 3 cases, respectively. In the remaining case, preoperative contrast-enhanced radiography revealed that the upper esophagus was higher than the clavicle; therefore, esophageal anastomosis with the cervical approach was performed.

In multistage surgery cases, the initial surgeries were thoracoscopic esophageal elongation for a long gap (four or more vertebrae), abdominal esophageal banding plus gastrostomy in one patient with ELBW (928 g), and thoracoscopic ligation of TEF plus gastrostomy in one patient with LBW (1590 g) and whose upper esophageal pouch was very fragile.

Gross type-D case

In the Gross type-D case, left thoracoscopic esophageal elongation for the right aortic arch with a vascular ring and thoracoscopic ligation of TEF were performed on day 0 of

Table 1 Characteristics of all patients ($n = 38$)

Gross classification	Type A	Type B		Type C		Type D	Type E
Total (cases)	3 (multistage)	1 (cervical approach)	26 (single stage)	3 (multistage)	1 (cervical approach)	1 (multistage)	3 (cervical approach)
Initial surgery	Elongation 2 Gastrostomy 1	NA	NA	TEF ligation + elongation 1 Banding + gastrostomy 1 TEF ligation 1	NA	TEF ligation + elongation 1	Cervical TEF ligation 2 Thoracoscopic TEF ligation 1
Timing of anastomosis (days of birth)	13 (11–34)	162	1 (0–3)	35 (7–106)	1	10	NA
Weight (g)	2550 (2319–3242)	5212	2588 (1678–3396)	2472 (2052–2878)	2196	2542	NA
Operative time (mm)	194 (183–214)	481	149 (73–245)	187 (145–243)	129	267	NA
Bleeding (ml)	4 (2–20)	101	1 (0–20)	5 (0–5)	1	5	NA
Hospitalization period (days)	121 (99–330)	32*	59 (18–300)	55 (53–282)	34	104	NA
Intraoperative complications	None	None	None	None	None	None	NA
Postoperative complications (cases)							
Minor leakage	1	0	2	0	0	0	0
Balloon dilatation for anastomotic stricture	1	0	4	1	1	0	0
Balloon dilatation tor congenial esophageal stenosis	0	0	4	0	0	0	0
Chylothorax	0	0	4	0	0	0	0
TEF recurrence	0	0	0	0	0	0	1
Gastroesophageal reflux disease	3	0	8	2	0	0	0
Mediastinitis after esophageal elongation	0	1	0	0	0	0	0

Single stage: thoracoscope single-stage anastomosis

Multistage: thoracoscope multistage anastomosis

Cervical approach: anastomosis or TEF ligation with cervical incision

Median (Minimum–maximum)

NA not applicable

GERD gastroesophageal reflux disease, *TEF* transesophageal fistula

*Re-hospitalization for anastomosis

birth followed by left thoracoscopic esophageal anastomosis on day 10 [6].

Gross type-E cases

Among the three Gross type-E cases, ligation of TEF was performed using the cervical approach in two cases. In the remaining case, thoracoscopic ligation of TEF was initially performed; however, TEF recurred subsequently, requiring second surgery using the cervical approach.

Single-stage surgery cases (Table 1)

In these cases, concurrent malformations included duodenal atresia plus anorectal malformation in two patients, anorectal malformation plus intra-abdominal testis in one, anorectal malformation in one, glucose phosphate isomerase (GPI)

deficiency plus Hirschsprung disease in one, and severe tracheomalacia in one. No concurrent malformations were observed in the remaining 20 patients.

With the exception of two patients who underwent tracheotomy and laryngo-tracheal separation for tracheomalacia and GPI deficiency, the length of hospital stay was 49 days (range 18–300 days).

Multistage surgery cases (Table 2)

Among multistage surgery cases, severe tracheomalacia was observed in four patients (three who underwent tracheotomy and one who underwent aortopexy), anorectal malformation in three, right aortic arch with a vascular ring in one, patent ductus arteriosus in two, and multiple malformations in one.

Complications

There were no cases of thoracotomies or intraoperative complications associated with thoracoscopic surgery. Although minor leakage was observed in three cases, it was conservatively resolved. There were three cases (9%) that required surgical treatment of postoperative chylothorax and seven cases of balloon dilatations (21%) for treating anastomotic stricture (Table 1). There was no case showing any thoracic deformities.

Chylothorax

Postoperative chylothorax was observed in 4 of 26 patients who underwent single-stage anastomosis. After we changed the operative procedure to ligation of the azygos vein without cauterization, chylothorax did not recur.

Gastroesophageal reflux disease (GERD)

Sliding hernia was observed in 5 of the 26 patients who underwent single-stage anastomosis. During 24-h pH monitoring, eight patients were diagnosed with GERD, including one with pepsinogen I deficiency, short esophagus, and multiple malformations. This patient underwent laryngo-tracheal separation and esophagogastric dissociation. The other seven patients were administered medications, and their symptoms resolved without fundoplication. In four of these seven patients who were more than 1 year old, no gastroesophageal reflux was observed with follow-up 24-h pH monitoring.

Of the eight patients who underwent multistage anastomosis, one with type-B disease who underwent cervical esophagogastric anastomosis was excluded. Five (3 type-A cases, 2 type-C cases) of the remaining seven patients were diagnosed with GERD based on 24-h pH monitoring and contrast-enhanced radiography. Sliding hernia was observed in two patients. All five patients were administered medications without fundoplication.

Anastomotic stricture and congenital esophageal stenosis (CES)

Among 26 type-C patients who underwent single-stage surgery, 8 required balloon dilatation, including 4 cases (15%) each for the treatment of anastomotic stricture and CES.

Among the 26 type-C cases, anastomotic stricture was observed under contrast-enhanced radiography in five cases immediately after surgery, and two of the five cases had stenosis that needed dilation. Two of the remaining 21 patients who had no anastomotic stricture immediately after surgery had a stricture that required dilation 1 year later.

Among four type-C patients who underwent multistage anastomosis, one (25%) required balloon dilatation. In that case, anastomotic stricture was not observed, but sliding hernia was observed under contrast-enhanced radiography within 1 year after surgery; however, anastomotic stricture was observed a year and a half after surgery. In the remaining three cases, anastomotic stricture was observed under contrast-enhanced radiography 1 month after surgery;

Table 2 Characteristics of seven patients who underwent multistage thoracoscopic esophago-esophageal anastomosis

	Case	Reason for multi-stage repair	First operation	Second operation	POD*
Type A	1	Long gap	Elongation	Thoracoscopic E-E anastomosis	12
	2	Long gap	Elongation	Thoracoscopic E-E anastomosis	8
	3	Long gap	Gastrostomy	Thoracoscopic E-E anastomosis	34
Type C	4	Long gap	Ligation of the TEF + Elongation	Thoracoscopic E-E anastomosis	6
	5	LBW (1590 g)	Ligation of the TEF + Gastrostomy	Thoracoscopic E-E anastomosis	34
	6	ELBW (928 g)	Banding of the abdominal esophagus + Gastrostomy	Thoracoscopic E-E anastomosis	105
Type D	7	Vascular ring (right aortic arch)	Ligation of the TEF + Elongation	Thoracoscopic E-E anastomosis	10

E-E esophago-esophageal, *EVLBW* extremely low birth weight, *LBW* low birth weight, *TEF* transesophageal fistula

*Days after first operation

however, it was no longer observed with no related symptoms such as stuck food at 1 year after surgery.

In addition, one of the three type-A cases required balloon dilatation. Of the seven patients who underwent multistage anastomosis, anastomotic stricture was observed in three patients requiring several dilatation sessions.

Discussion

Thoracoscopic repair of EA is associated with excellent results and can be performed with a good visual field of the cranial and diaphragm sides. In patients with a long gap, the procedure has been reported to enable anastomosis, while other reports indicate no difference between thoracoscopy and open surgery in terms of anastomotic failure and postoperative anastomotic stricture [7].

A few reports indicate that thoracic deformation occurs to a lesser extent with thoracoscopic repair than with thoracotomy; thus, thoracoscopic repair is highly advantageous esthetically in the short and long term [7–12]. No cases of thoracic deformity were observed in our series.

A shortcoming of thoracoscopic repair of EA is that the operation is technically difficult [13]. A few reports have indicated that patients with LBW and long gaps should be excluded from these indications [14, 15]. Patients with LBW are treated in multiple stages; first, esophageal banding or TEF resection is performed alone; then, we wait for the patient to grow, and finally, we perform thoracoscopic anastomosis. This anastomosis is not difficult to perform; therefore, the strategy is acceptable. Patients with a long gap or vascular ring were treated by thoracoscopic elongation in the first stage. With this multistage treatment, thoracoscopic esophago-esophageal anastomosis can be performed in all except one patient who exhibited mediastinitis after the first operation, precluding anastomosis at the second stage. In cases of Gross type-A, we decided to perform elongation before the initial surgery. Now, we attempt to perform only gastrostomy during the initial surgery. In cases of long gap with more than 5 vertebrae or a very fragile upper esophagus, elongation was performed based on the intraoperative decision taken. After 6–12 days, we performed thoracoscopic delayed primary esophago-esophageal anastomosis. Conversion to open surgery occurs in 5–15% cases, with the inability to perform anastomosis because of strong tension, bleeding, and poor visualization [13–18]. However, in our case series, all procedures were successfully completed without conversion to open surgery. Conversion and complications may have been avoided by limiting the number of operators in cases with fragile tissues and weighing < 2000 g. In our anastomosis procedure, the first two sutures were started from the mucosa of the distal esophagus using a reverse needle to ensure that the mucous was sutured. We also ensured that the seams were not too fine, and we stitched around the entire circumference of the esophagus using six needles. Our anastomosis procedure may also have contributed to the results. In our series, no patient had severe heart disease. Based on these results, thoracoscopic esophageal anastomosis should be possible for all patients, regardless of the disease type and patient condition, except in cases of unsuitable hemodynamics, such as heart disease or limited populations with type-E surgery or with the end of the upper esophagus located higher than the clavicle, necessitating the cervical approach (Fig. 1).

Following single-stage anastomosis, GERD occurred at a rate close to 70%; however, it was resolved with conservative follow-up as well as observation. Although anastomotic stricture was only observed in the early period in approximately 10% of the patients, it should be noted that the stricture can appear at more than 1 year postoperatively.

Multistage thoracoscopic radical surgery is indicated for patients with LBW, a long gap, and a vascular ring. GERD, including postoperative esophageal hiatal hernia, occurred in approximately 80% of the patients, and anastomotic stricture

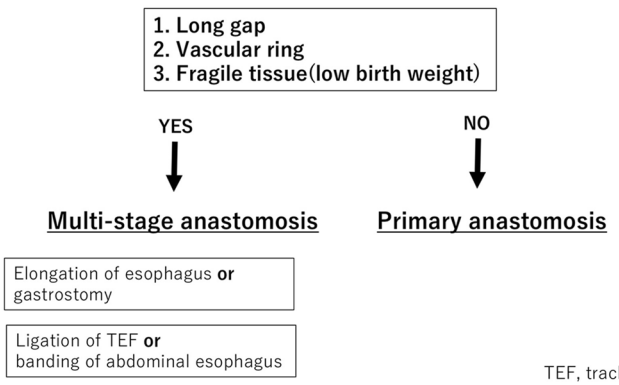

Fig. 1. Our strategies for thoracoscopic repair of esophageal atresia. *TEF* transesophageal fistula

was observed in the early stage in approximately 50% of the patients. Therefore, the appropriate use of medication, contrast-enhanced radiography, pH monitoring, and a thorough, long-term observation should be performed. Multistage thoracoscopic surgery caused multiple postoperative complications compared to the single-stage operation. In particular, one patient with a long gap had many postoperative complications. A long gap indicates a short esophagus. Our elongation method is good but is not sufficient to elongate a native short esophagus. Anastomosis of a short esophagus might cause sliding hernia, anastomotic stricture, and other complications. We must therefore identify and pursue more effective and innovative methods to elongate a short esophagus.

According to our thoracoscopic repair of EA, the outcome of our strategy is summarized Fig. 1. Based on this figure, we intend to carry out future repair of EA.

Conclusions

Although the limited number of cases and short-term observation period prevented the investigation of long-term outcomes, thoracoscopic radical repair can be indicated for most types and conditions of EA in patients with stable hemodynamics. The number of operators should be limited in cases with fragile tissues and weighing < 2000 g. Postoperative management should be performed with care, considering that chylothorax, GERD, and anastomotic stricture might occur.

Compliance with ethical standards

Conflict of interest All the authors have no conflicts of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Research involving human participants All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee (ethics review board at Nagoya University Graduate School of Medicine: Approval No. 2016-0499).

Informed consent Informed consent was obtained from all individual participants included in the study.

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