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Experience in the treatment of type C congenital esophageal atresia using a staged approach

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Abstract

Background In select patients with type C esophageal atresia, primary anastomosis is not appropriate and a staged approach is required. We aim to summarize our experience in the management of type C EA using a staged approach.

Methods A retrospective chart-review of patients with type C EA admitted to Beijing Children's Hospital between July 2020 to October 2023 were conducted. Those diagnosed with type C EA who were not amendable to primary anastomosis were included for analysis. Clinical information was recorded, and follow- up was performed.

Results Seven (five boys) patients with type C EA who received staged repair were included in the study. Initial surgeries included thoracotomy and thoracoscopy. 71% (5/7) patient had complications after the initial surgery, including pyopneumothorax, pneumonia, recurrent tracheoesophageal fistula (rTEF), and anastomotic leak. Esophageal elongation techniques were applied in 3 patients. All delayed anastomosis were performed thoracoscopically, except for in one case where spontaneous fistulization occurred and no anastomosis were necessary. Complications after delayed anastomosis included recurrent esophageal pulmonary fistula (rEPF) in 50% (3/6), anastomotic leak in 33% (2/6), and esophageal stricture in all (6/6) patients. After a median follow-up of 14 months (range: 2–24), all patients were in generally good condition.

Conclusion Primary operations should be kept simple and minimal in patients diagnosed with type C EA who are not appropriate for primary anastomosis. Internal traction is an effective method that allows for subsequent anastomosis. Intraoperative indocyanine green fluorescence can aid in fistula determination and anastomosis.

Keywords Esophageal atresia, Staged surgery, Thoracoscopy

Introduction

Congenital esophageal atresia (EA) with or without tracheoesophageal fistula (TEF) is a serious and life-threatening anomaly of the gastrointestinal tract with an estimated incidence of 1/2500 ~ 1/4500 [1]. According to Gross classification, type C is the most common, characterized with a distal TEF [2]. Treatment of type C EA is frequently done through primary anastomosis. However, in select patients, primary anastomosis is not feasible and a staged approach is required. These cases may include long gap atresia [2, 3], and patients with poor general or

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local conditions [4, 5]. A recent report of 38 EA infants highlighted the importance of preoperative consideration of multi-stage surgery for better outcomes of these complex cases [4]. Yet, discussion on the surgical techniques for staged operation of type C EA is lacking and largely dependent on the surgeon's experience [6]. In this paper, we present seven cases of staged repair of type C EA patients, summarized their clinical characteristics and report our experience in the surgical management of these complex cases.

Material and methods

Patients

A retrospective chart-review of patients with type C EA admitted to Beijing Children's Hospital from July 2020 to October 2023 were conducted. Those diagnosed with type C EA who were deemed not amendable to primary anastomosis were included for analysis. Patients' demographic and clinical information, operative management, complications, outcomes and follow-up status were reviewed. Follow-up was performed in the form of outpatient visits or telephone interviews.

All studies were conducted in accordance with the Declaration of Helsinki. Due to the retrospective nature of this study, the need for informed consent was waived by the Medical Ethics Committee of Beijing Children's Hospital (2019-K-333). This study does not include participants above 16 years of age.

Preoperative assessment

Preoperative esophagography is ordered as an initial assessment, followed by insertion of bougies orally and via gastrostomy to assess if the two ends overlap. Overlapping esophageal ends warrants anastomosis. Bronchoscopy is also performed to assess for development of the trachea and to rule out recurrent TEF.

Surgical procedure

In a staged approach of type C EA, the first surgery entails ligation of the TEF and pleural adhesiolysis where necessary. Details of the surgical procedures to correct TEF under thoracoscopy were reported in our prior study [7, 8]. After general anesthesia and tracheal intubation, an endoscope was passed down and 0.25 ml indocyanine green (ICG) (2.5 mg/ml) was sprayed at fistula to assist in precise dissection later. The patient was then placed in the left prone position and properly draped. A 5 mm trocar was placed in the fourth intercostal space of the right scapular line, and two 3 mm trocars were placed in the third and sixth intercostal spaces of the right midaxillary line. The pressure inside the chest was maintained at 6–8 mmHg. After meticulous dissection to release the esophagus from the trachea, the fistula was revealed under thoracoscopy, ligated using a 5–0 polydioxanone suture and transected. Loose tissue of the prevertebral fascia may be placed between the two ends to prevent recurrence. The two pouches of the esophagus may be pulled with moderate tension to assess for feasibility of a primary anastomosis: If the overlap between the two ends was less than 0.5 ~ 1 cm, a primary anastomosis was deemed inappropriate (see Fig. 1). In these cases, the proximal pouch was left unopened and the distal pouch was sewed using 5–0 interrupted absorbable suture. The anterior and posterior wall of both pouches were fixed onto the prevertebral fascia under moderate tension. Traction force was carefully controlled so as to prevent tearing through the esophagus causing leakage and subsequent infection. Myotomies were not performed.

The second phase of the repair was aimed to establish anastomosis. After general anesthesia and tracheal intubation, an endoscope was passed down and ICG was sprayed on the esophageal pouch to assist in anastomosis later (see Fig. 2). Three trocars were placed in the above stated fashion. After adequate adhesiolysis, the surgeon switched to fluorescence mode to aid in finding both ends

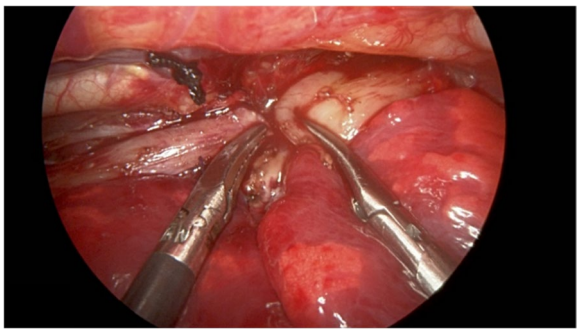

Fig. 1 Case 7 Intraoperative picture: the two ends of the esophagus cannot overlap under tension

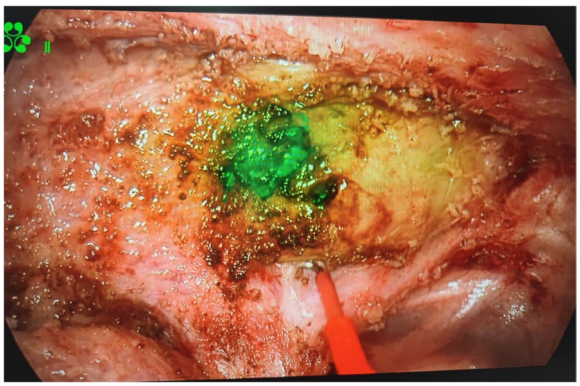

Fig. 2 Intraoperative imaging of ICG application revealing the proximal esophageal pouch

of the esophagus. Adhesions were carefully separated to mobilize the esophagus: the proximal end to the thoracic inlet and the distal end to the diaphragm. The two ends were pulled under moderate tension so that they overlap for about 1 cm (see Fig. 3), and perfusion of esophagus were assessed. The proximal blind pouch was opened, scarring on the two pouches were removed. The posterior wall of the esophagus was sutured first using five 5–0 PDSII interrupted sutures, then a French 6 gastric tube was inserted under direct vision. The anterior wall was closed with five interrupted sutures. After checking for leakage, perfusion and adequate hemostasis, a chest tube was placed on the right side.

Results

Patient characteristics

Table 1 presents the general characteristics of seven (five boys and two girls) type C EA patients included in the study. All patients received staged surgical repair. Except for Case 4 and 7, whose primary operation were done at our center, all other patients underwent primary operation at other facilities. The median gestational age was 38 weeks (range: 29–40); the median birth weight was 3.03 kg (range:1.37–3.27); the median age at initial visit at our center was 82 days (range:1–230), and the median weight at initial visit at our center was 3.6 kg (range:1.44- 8.5). Two patients were born prematurely at less than 37 gestational weeks. One patient had concomitant congenital anal atresia with recto-urethral fistula, and two patients with congenital heart disease.

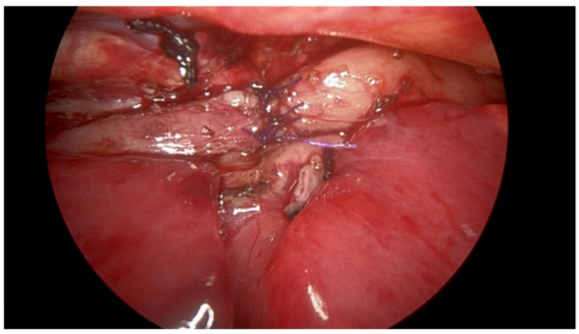

Fig. 3 Case 7 Intraoperative picture: the two ends of the esophagus sutured onto the prevertebral fascia at the level of T3 using 5-0PDS suture without myotomy

Treatment process

Perioperative information is shown in Table 2. Initial surgeries included both thoracotomy and thoracoscopy. 71% ($n=5/7$) patients had complications after the initial surgery, including pyopneumothorax ($n=1/7$), pneumonia ($n=1/7$), recurrent tracheoesophageal fistula (rTEF) ($n=2/7$), and anastomotic leak ($n=1/7$). Gap length was measured during repair of EA/TEF at our center thoracoscopically. Esophageal elongation technique was performed in 3 patients, including internal traction and bougienage stretching in 2 (Case 1 and 2) and bougienage stretching alone in 1. All delayed anastomosis were performed thoracoscopically, except for in one case where spontaneous fistulization occurred and no anastomosis were necessary. Complications after delayed anastomosis included recurrent esophageal pulmonary fistula (rEPF) in 50% ($n=3/6$), anastomotic leak in 33% ($n=2/6$), and esophageal stricture in all ($n=6/6$) patients.

Post-operative care

Chest tubes were inserted in all cases and all were transferred to PICU/NICU after surgery. All patients were given sedation, analgesia, antibiotics and ventilatory support. The median duration on ventilatory support was 9 days (range: 7- 21 days). Patients were transferred to ward after weaning off ventilator and stabilized.

Table 1 General characteristics

Case	Sex	Gestational age (Weeks)	Birth Weight	Age at initial visit[a] (Day)	Weight at initial visit*(kg)	Comorbidity	Follow up since delayed anastomosis[b] (month)
1	male	38	2.7	85	3.6	CAA with RUF	24
2	male	38^{+6}	3.1	82	4.0	PDA, ASD	14
3	male	38	3.03	230	8.5	NA	25
4	male	40^{+4}	3.27	1.5	3.2	NA	39
5	male	37^{+2}	3.1	113	5.3	NA	3
6	female	36	2.2	34	2.8	PDA	8
7	female	29^{+6}	1.37	1	1.4	NA	1

Annotation: *CAA* congenital anal atresia, *RUF* recto-urethral fistula, *PDA* patent ductus arteriosus, *ASD* atrial septal defect, *NA* not applicable

* measured at first visit at our center

[a] first visit at our center

[b] follow-up as of December 2023

Table 2 Perioperative information

Case	Initial repair of EA/TEF	Complications after initial repair	Gap length* (cm)	Esophageal elongation	Delayed anastomosis	Complications after delayed anastomosis			Ventilatory support (day)
						Fistula	Anastomotic	Stricture (number of dilations)	
1	Thoracotomy + myotomy	pyopneumothorax	6	IT; bougienage	Thoracoscopy	$rEPF_2$	-	+ (48)	10
2	Thoracotomy	$rTEF_2$	3.5	IT; bougienage	Thoracoscopy	-	-	+ (1)	7
3	Thoracoscopy	$rTEF_2$	4	NA	Thoracoscopy	$rEPF_2$	-	+ (39)	9
4	Thoracoscopy	-	5	NA	Thoracoscopy	-	leak	+ (12)	13
5	Thoracoscopy	anastomotic leak	3[1]	NA	Thoracoscopy	-	-	+ (2)	8
6	Thoracotomy	pneumonia	3[0.5]	bougienage	Thoracoscopy	$rEPF_1$	leak	+ (13)	8
7	Thoracoscopy	NA	2.5	IT	Spontaneous fistulization	NA	NA	NA	21

Annotation: *rTEF* stands for recurrent tracheoesophageal fistula, *rEPF* stands for recurrent esophageal pulmonary fistula, *IT* stands for internal traction, *NA* stands for not applicable

+ represents a positive diagnosis;—represents a negative diagnosis; number in footnotes represents total occurrences; *measured during initial repair of EA/TEF at our center thoracoscopically; [] overlapping length measured under tension

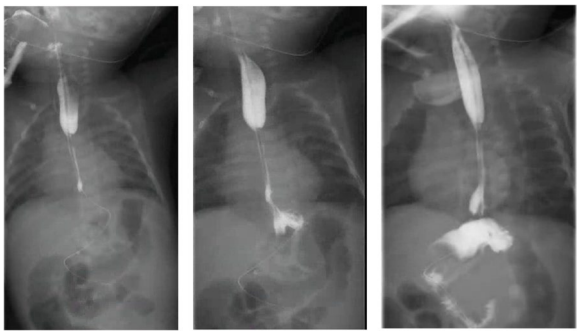

Fig. 4 Esophagogram of case 7 at three months reveals spontaneous fistulization of the two ends of the esophagus without leakage

Esophagography was performed routinely 2 weeks after operation to rule out anastomotic complications.

Follow-up

Median follow-up was 14 months (range: 2- 24 months). All patients were taking oral feeds and gaining weight. One patient developed anastomotic leakage and was managed conservatively using naso-jejunal feeding tube and an upper esophageal suction with good results. Three children are currently receiving periodic endoscopic balloon dilatation for esophageal strictures. At the time of writing, Patient 7 is three months post-operation and an esophagogram revealed spontaneous fistulization of both ends of the esophagus without signs of leakage (see Fig. 4). All other children showed no other complications at regular follow-up and esophagography.

Discussion

Our study presents seven cases of staged repair for type C EA in patients with complex surgical or general conditions, including poor thoracic status, long gap length, and very low birth weight. These are also the same challenges faced by many pediatric surgeons. Based on our experience, we consider gap length to be the most critical determinant for performing a staged repair. Despite significant improvements in the management of congenital EA, long gap esophageal atresia (LGEA) is rather a unique entity. Efforts have been made to define and standardize the management of LGEA, yet the precise definition, gap length determination, and surgical approach to LGEA is still controversial [9]. Many agree that the inability to achieve primary end-to-end anastomosis after adequate mobilization as working definition of LGEA [2, 9–11]. This is also the definition we chose for the discussion of this paper.

Accurate determination of the gap length between the proximal and distal pouch of the esophagus prior to operation is often difficult, and there are many variations reported in the literature [9, 10, 12]. At our center, esophagography is routinely performed pre-operatively for its vital role in determining location and size of the proximal pouch, while bronchoscopy may aid in detecting tracheoesophageal fistulas and location of the distal esophagus. Sometimes a 3D reconstruction CT is helpful in determining gap length between the two ends of the esophagus. A staged repair may be chosen in cases where the proximal pouch ends above the level of T2, or distal TEF opens at the carina [13].

In regards to the operative approach of a staged repair, we advocate for a simple and minimal primary procedure. Specifically, myotomies of the proximal pouch should be avoided, as evidenced by our experience in the management of Case 1. In this case the patient underwent an initial myotomy of the proximal esophagus during primary surgery at another institution. Postoperatively, the patient developed an anastomotic leakage that led to a life-threatening pyopneumothorax. Although the child was managed conservatively using a chest tube and an upper esophageal suctioning, the infection caused extensive adhesion in the thorax and posed significant challenges to subsequent operations. Our view aligns with a recent publication by Galazka, Skinder [4], who similarly advocated for simpler primary surgery by performing only fistula ligation or internal traction placement.

There are various surgical techniques to repair LGEA with no clear consensus on the preferred method. Esophageal replacement was once a popular technique, most commonly gastric transposition or colonic interposition [9]. However, its use as first-line therapy has been limited due to variable short- and long-term complications, including anastomotic leak, stricture, GERD, gastrointestinal and respiratory symptoms [14–17]. It is now agreed that there is no ideal substitute for the native esophagus, hence efforts at preserving the native esophagus has grown to be the ideal approach [18, 19]. A range of esophageal lengthening techniques have been reported to allow for primary anastomosis, including natural growth, esophageal myotomy (both circular and spiral myotomy), Foker and Kimura's technique and their variations [20–22]. The most recent adaptation from the Foker's technique was described by Dr. Patkowski, who introduced an internal traction suture system that creates an evenly distributed traction force on the esophagus only [23, 24]. These procedures are technically demanding, requiring substantial surgical expertise, and carry a risk of severe complications, including anastomotic leak, esophageal diverticulum and rTEF, therefore their uses are not as popularized [25, 26]. In recent years, elongation by bougienage stretching technique has been described by Sun, Pan [27] that has resulted in optimal results. We recommended that if primary reconstruction was not possible, the initial surgery ought to be simple and operation time kept short. Esophageal elongation techniques, such as external traction or Kimura's technique, should be avoided to prevent severe complications. In case of a confirmed type C EA not amendable to primary repair, it is recommended to treat only the TEF and not mobilize or perform internal traction unless performed in experienced hands. In our series, two patients (Case 1 and 2) underwent internal traction which had permitted for growth and improved elasticity of the esophagus that were essential for anastomosis. It was noted that the outcomes associated with internal traction was less favorable in the presence of extensive and severe adhesions, as demonstrated in Case 1. These adhesions are likely a consequence of multiple prior surgeries and infections, further supporting the principle of a simple primary procedure.

Timing of delayed anastomosis is very important. Growth of the esophagus is faster than somatic growth due to swallowing and gastric reflux 8 to 12 weeks after birth, therefore the ideal timing for delayed reconstruction is around 12 weeks after birth [28]. While awaiting surgery, a suction catheter placed in the upper pouch that allows for continuous drainage of saliva is vital in prevention of aspiration pneumonia and excessive enlargement of the proximal pouch. Case 3, aged 7 months and 16 days at the time of admission, was not placed an upper pouch suction tube at other hospital, and the proximal esophageal pouch was evidently dilated with a thin muscularis as noted intraoperatively. Such condition increased the risk of anastomotic leak.

Application of ICG fluorescence has gradually gained popularity in pediatric surgery in recent years [8, 29, 30]. Its use in pediatric gastrointestinal surgeries were primarily for assessment of tissue perfusion to prevent complications and precise incision [31]. We have previously published our results of application of ICG fluorescence in esophageal pulmonary fistula after esophageal atresia repair [8]. These operations presented special challenges as patients had received multiple surgeries previously and resulted in extensive adhesion and altered anatomical positions, therefore precisely locating the esophagus was essential to repair. 0.5 ml ICG (2.5 mg/ml) was sprayed onto both ends of the esophageal pouches under the assistance of endoscope preoperatively. Under fluorescence mode, the proximal and distal esophageal ends were clearly observable after adhesiolysis, providing a safe and convenient working space for the surgeon. In this current series, ICG fluorescence was applied in all patients for precise location, assessment of gap length and safe-check for anastomotic leakage.

Rate of stricture is high after staged repair in this series. All seven patients required dilations after anastomosis. This is attributable to dissociation of esophagus that may lead to relatively poor perfusion, higher tension at the anastomosis, and gastric reflux after gastrostomy. This complication can be safely and effectively managed with endoscopic balloon dilatation [32, 33] in most cases. This is consistent with the clinical outcome observed in the current series as well. It is worthy of further discussion of Case 1 and 3 in this series who required multiple dilations. First, extensive scarring was noted in both cases during thoracoscopy, particularly on the distal

esophagus, which were likely resulted from inflammatory edema caused by excessive dissection or failed attempt at anastomosis during the initial procedure performed at other institutions. Second, the esophageal gap lengths in both cases were substantial (Case 1: 6 cm; Case 3: 4 cm), consequently increased tension at the anastomotic site. These factors contributed to significant post-anastomotic scarring, that required multiple dilatations. Surgical resection of the strictures was proposed; however, the parents declined this procedure. These complex cases provide further evidence of the need for a simplified initial surgery when primary anastomosis was not feasible. At follow-up, all patients were able to tolerate oral feeds after dilatation without the need for resection of the stricture. Esophageal pulmonary fistula is a special type of complication after EA repair, which has rarely been reported in the literature. We have published our management of esophageal pulmonary fistula after EA repair in another study in detail [8].

Despite the tremendous improvements in the prognosis of EA patients in general, the surgical treatment of preterm and very low birthweight infants is still a challenge to surgeons. Current reports of this delicate population are often of small series [34–36]. Surgical risks, surgical approach, management of postoperative complications and prognosis are limited and more reports are needed. In this series, we reported a very low birthweight (1.44 kg) preterm baby, Case 7, who underwent operative treatment 6 days after birth. Due to her small bodyweight, a longer gap length (2.5 cm), and the higher risk of anastomotic leak, we only ligated and severed the TEF and performed internal traction and fixation of both ends of the esophagus on the prevertebral fascia at the level of T3. A gastrostomy was created for feeding. Both ends of the esophagus fistulized spontaneously at three months without signs of complications. Stringel, Lawrence [37] reported similar observation of spontaneous fistulization after suturing the two ends of the esophagus. As this is the only case in this series, more reports and prospective studies are needed to establish treatment approach to this delicate patient population. The significance of this case also lies in the observation of spontaneous growth and fistulization after internal traction and might provide new insights into the treatment of EA in preterm and very low birthweight babies.

We have derived the following surgical principles from our experience in the management of staged repair of type C EA:

1. Adequate dissection, ligation and transection of the TEF, then suture and fix the distal esophageal pouch onto the prevertebral fascia avoiding the original TEF site to prevent recurrence;
2. Without performing myotomy, the proximal pouch is closed with 3 to 4 mattress sutures using 5-0prolene suture without piercing through the lumen of the esophagus. The proximal pouch is mobilized, brought close to the distal esophagus under adequate tension, and fixed onto the prevertebral fascia;
3. Cervical esophagostomy is not necessary. However, a suction tube inserted via the nostril is essential for continuous drainage of the upper pouch;
4. Delayed anastomosis should be performed at least 1 months from the initial surgery when the baby weigh at least 3 kg;
5. Internal traction techniques can promote spontaneous growth of the esophagus and enhance elasticity that allow for later anastomosis.

There were some limitations to this study. The retrospective nature of this study has its intrinsic flaws. Due to the rarity of type C esophageal atresia needing staged surgery, only seven patients were identified. The clinical presentation, diagnoses, and treatment of the condition can only be discussed preliminarily. Considering the probability of long-term recurrence, further observation, follow-up and prospective studies are needed.

Conclusion

To conclude, operations should be kept simple and minimal in patients diagnosed with type C EA who are not appropriate for primary anastomosis. Myotomy of the esophageal pouch should be avoided in these cases to prevent risks of post-operative leak and pyopneumothorax. Moreover, excessive manipulation and mobilization of esophagus may lead to poor perfusion and altered anatomical position, also increasing difficulties to subsequent operations and risks of post-operative stricture. Internal traction techniques are effective at promoting growth of the esophagus that eventually allow for anastomosis. ICG fluorescence during repair can help precisely determine location of the fistula.

Abbreviations

EA	Esophageal atresia
rTEF	Recurrent tracheoesophageal fistula
ICG	Indocyanine green

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Clinical trial number

Not applicable.

Authors' contributions

Y.Z.,S.T., and A.W. drafted the manuscript text and all tables. S. L., J.L, D.W., K. Hua, Y.Gu critically reviewed and revised the manuscript and tables. Y.Z. and J.H contributed to the funding, resources, pictures, and supervision of this study.

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Data availability
Data is provided within the manuscript.

Declarations

Ethics approval and consent to participate
Information obtained from the data of the study participants is kept confidential. To ensure confidentiality, the names of study participants were not included in the data. All studies were conducted in accordance with the Declaration of Helsinki. Due to the retrospective nature of this study, the need for informed consent was waived by the Medical Ethics Committee of Beijing Children's Hospital (2019-K-333). This study does not include participants above 16 years of age.

Consent for publication
Not applicable.

Competing interests
The authors declare no competing interests.

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