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Revisional surgery for recurrent tracheoesophageal fistula and anastomotic complications after repair of esophageal atresia in 258 infants

Antti I. Koivusalo *, Mikko P. Pakarinen, Harry G. Lindahl, Risto J. Rintala

Children's Hospital, Section of Paediatric Surgery, University of Helsinki, Helsinki, Finland

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ABSTRACT

Aim: We assessed the occurrence and outcome of major reoperations following repair of esophageal atresia with or without tracheoesophageal fistula (TOF). Major outcome measures were survival, preservation of native esophagus, and long–term esophageal function.
Methods: Hospital charts of 258 consecutive patients treated for esophageal atresia from 1980 to 2013 were reviewed.
Results: Forty-two (16%) patients required a total of 57 reoperations after primary repair ($n = 37$) or esophageal reconstruction ($n = 5$). The indications were anastomotic leakage ($n = 17$), anastomotic rupture after endoscopic dilatation ($n = 5$), recurrent tracheoesophageal fistula (TOF) ($n = 12$), undiagnosed proximal TOF ($n = 3$), recalcitrant anastomotic stricture ($n = 11$, primary anastomosis 9, reconstruction 2), undetected proximal fistula ($n = 3$), and inadvertently perforated jejunal graft ($n = 1$). Anastomotic leakage and rupture after dilatation were treated with rethoracotomy and suture and recurrent or undetected TOF by open repair. Strictures not manageable with repeated dilatations were resected and esophageal ends reanastomosed ($n = 10$) or bridged with jejunum graft ($n = 1$). Five (12%) patients required further reoperations, two after recurrent TEF (reocclusion $n = 1$, reconstruction with gastric tube $n = 1$), two after stricture operations (re-resection $n = 1$, resuture after leakage $n = 1$), and one after recurrent dilatation-related rupture. Mortality was 4/42 (10%). Two patients died of recurred leakage or TOF and two of unrelated cause. Of 38 survivors, 35 retained their native or initially reconstructed esophagus, and 3 had secondary reconstruction. After a median follow-up of 23 (range 0.6–32) years, 35 (95%) patients have acceptable esophageal function. Three patients remained dependent on gastrostomy feedings.
Conclusion: Anastomotic and TOF complications required a substantial number of reoperations, including esophageal reconstructions. Over 90% of the patients survived with a functioning native or reconstructed esophagus.

The repair of an esophageal atresia (OA) is usually followed by an excellent outcome. Still, the repair may fail or become complicated resulting in morbidity and occasionally mortality. In our previous review from Helsinki [1], leakage of the esophageal anastomosis occurred in 7%, recalcitrant anastomotic stricture occurred in 6% and a recurrent tracheoesophageal fistula (TOF) with or without a primarily undiagnosed proximal TOF (type D OA) occurred in 6%–10% of infants. All complications expose a neonate to infection and problems of nutrition and respiratory dysfunction. Reoperations are often complex and require experience and judicious timing. A variety of reconstructive procedures may be required to regain esophageal continuity [2].

The aim of this study was to describe the outcome after revisional esophageal surgery for the complications following repair of OA in a cohort of 42 children extracted from our series managed in our institution between 1980 and 2013.

1. Patients and methods

1.1. Study design

This was a retrospective case note review and followed institutional ethical committee approval. The case-notes were reviewed of 258 consecutive infants with OA admitted into Children's Hospital of Helsinki University Hospital between 1980 and 2013. We included all infants who underwent reoperative surgery for anastomotic leakage, anastomotic stricture, recurrent TOF or TOF which had remained undiagnosed during the primary repair. We excluded operations for gastroesophageal reflux (GOR), operations for tracheomalacia and other associated disorders, endoscopic procedures and stents. Follow-up included radiological imaging, endoscopy findings and growth outcomes.

The initial defect was classified according to Gross classification [3] i.e. Type A (OA without TOF), Type B (proximal TOF only), Type C (OA and distal TOF), Type D (OA and both proximal and distal TOF) and Type E (isolated TOF) depending on the surgical anatomy. Long-gap OA was defined by location of the distal TOF and judgement of the operating surgeon.

* Corresponding author at: Children's Hospital, Stenbackinkatu 11, 00290, PO BOX 281, Helsinki, Finland. Tel.: +358 504272512.
E-mail address: antti.koivusalo@hus.fi (A.I. Koivusalo).

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Compared with our earlier report [1] this study had a larger initial cohort of patients (258 patients from 1980 to 2013 compared with 130 patients from 1991 to 2012) and concentrates on surgical management of complications of esophageal anastomosis.

1.2. Principles of surgical management

Our techniques of repair of OA in our institution have been described in detail elsewhere [1]. Rigid bronchoscopy is routinely performed prior to the repair and the surgical approach in for the majority was a right transpleural thoracotomy. Recently an extrapleural approach was also used. The thoracoscopic approach was used in three infants only. Infants who were not amenable for primary end-to-end repair were managed with feeding gastrostomy and upper pouch suction drainage or cervical esophagostomy. The gap between upper and lower esophageal pouches was measured before definitive repair by double flexible endoscopy under fluoroscopic control. Delayed primary closure or esophageal reconstruction was scheduled at a minimum age of two months. Methods of esophageal reconstruction were reversed gastric tube (RGT) [4] or pedicled jejunal interposition graft (JIG) [5].

1.3. Treatment of postoperative complications

1.3.1. Anastomotic leakage

Postoperative anastomotic leakage was suspected if the patient had recurring pneumothorax and/or leak of air, saliva and gastric contents from the chest drain. Confirmation of the leakage was performed by chest x-ray with oral contrast. Primary treatment of anastomotic leakage included chest drainage, antibiotics and nutrition via nasogastric tube with parenteral nutrition support. If the leakage could not be controlled with these measures and the leakage was recent without signs of severe infection rethoracotomy and resuture was performed. If possible the sutured area was covered with a flap of vascularized tissue (pleura, pericardium, intercostal muscle). After one week healing was confirmed with contrast esophagogram and oral feeding started. Anastomotic dilatations were started 2–3 weeks thereafter. Cervical esophagostomy, suture closure of the distal esophagus and gastrostomy were performed in those with severe pleural contamination or wide anastomotic defect. Reconstruction of esophageal continuity was scheduled after 3 to 12 months.

Routine repeated postoperative dilatations were performed until the anastomotic width was 10 mm until 2001. Since then, dilatations have been performed selectively. Major dilatation associated anastomotic ruptures were treated by rethoracotomy and resuture as for anastomotic leakage.

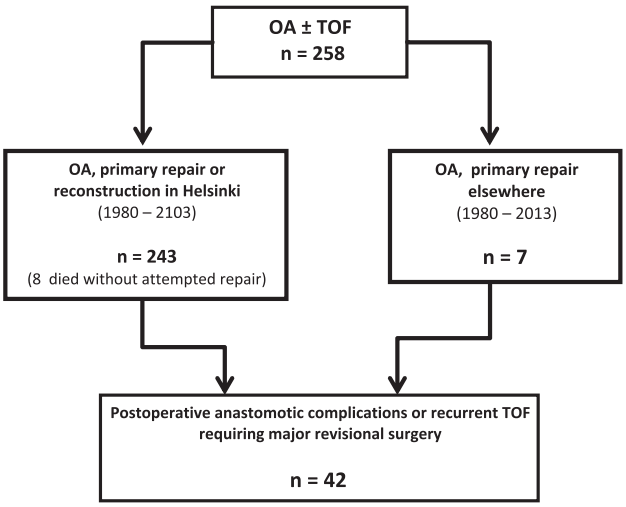

Fig. 1. Origin of patients with major anastomotic complications ($n = 42$).

1.3.2. Recurrent TOF

Recurrent TOF or undiagnosed second TOF was suspected in infants with feeding difficulties and recurrent chest infections. A number of methods were used for diagnosing recurrent TOF. These included rigid bronchoscopy with intubation of the pit of the initial TOF with a ureteral catheter or by injection of dilute methylene blue; contrast injected into a nasogastric tube slowly withdrawn during injection; or filling the esophagus with water during gastroscopy during manual continuous positive airway pressure (CPAP) ventilation and watching for a jet of bubbles.

If recurrent or undiagnosed TOF allowed intubation with a catheter, the catheter introduced through the fistula from the trachea was retrieved from the mouth with gastroscope and the looped catheter was used as a guide for intraoperative identification.

The surgical approach was transpleural through right lateral thoracotomy or right cervical incision for high fistulas. The fistulous openings were closed by suture and an attempt to interpose vascularized pericardium, pleura or a strip of intercostal or neck muscle. Anastomotic resection and reanastomosis was considered in those with concomitant stricture.

1.3.3. Recalcitrant anastomotic stricture

Primary treatment of recalcitrant anastomotic stricture was serial endoscopic balloon dilatations, recently with topical mitomycin application [6] coupled with medical and surgical control of GOR. Esophageal resection was considered if a dilatator guide wire could not be passed through the stricture, the stricture failed to yield to dilatations or if, after numerous dilatations, the esophageal lumen repeatedly occluded and caused dysphagia or respiratory problems. Esophageal continuity was reestablished with end-to-end anastomosis or, in long strictures or if substantial length of esophagus was lost, RGT or JIG.

Data are quoted as median (range). Statistical comparisons were made with StatView® 512 computer programme (Brain Power, Calabasas CA, USA). Risk ratios for different outcome variables were estimated using univariate logistic regression analysis. P values of <0.05 were considered statistically significant.

2. Results

A total of 258 infants with OA ± TOF were treated from 1980 to 2013. Complications included early anastomotic leakage ($n = 20$, 8%), anastomotic rupture after endoscopic dilatation ($n = 5$, 2%), recurrent TOF ($n = 14$, 5%), undiagnosed proximal TOF ($n = 3$, 1%), recalcitrant anastomotic stricture ($n = 11$, 4%) and others ($n = 1$).

Revisional surgery was performed in a total of 42 patients and included 35 (14%) of our 251 children with primary repair and 7 children referred from other hospitals. (Fig. 1) their median birth weight was 2.6 (0.9–4.8) kg and gestational age 38 (30–42) weeks. Birth weight was <1500 g in 4 infants. Types of OA were as follows: A ($n = 7$), C ($n = 30$), D ($n = 3$), and E ($n = 2$). According to the Spitz classification [7], 33 (79%) patients belonged to Group I (no major cardiac anomalies and birth weight ≥1500 g) and 9 (21%) to Group II (major cardiac anomaly or birth weight <1500 g) and none to group III (major cardiac anomaly and birth weight <1500 g). Eighteen (43%) had an associated congenital disease including significant heart disease ($n = 7$), congenital lobar emphysema ($n = 1$), anorectal malformation ($n = 9$), duodenal atresia ($n = 4$), choanal atresia ($n = 2$), omphalocele ($n = 1$), urogenital anomaly ($n = 5$), anomaly of limbs or vertebrae ($n = 5$), Down's syndrome ($n = 3$), Feingold's syndrome ($n = 1$) and CHARGE association ($n = 2$). Fifteen (36%) of the 42 patients required antireflux surgery for symptomatic GOR within the first year of life and eight (19%) underwent aortopexy for significant tracheomalacia within the first three months of life.

Fifty-seven major revisional operations were performed in 42 children [following primary end-to end repair ($n = 37$) and esophageal reconstruction ($n = 5$)]. Two or more operations were performed in 11