Journal of Pediatric Surgery 53 (2018) 925–928

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Repair of congenital esophageal atresia with tracheoesophageal fistula repair in Ontario over the last 20 years: Volume and outcomes

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ARTICLE INFO

Article history:
Received 25 January 2018
Accepted 1 February 2018

Key words:
Esophageal atresia repair
Tracheoesophageal fistula repair
Outcomes
Volume

ABSTRACT

Background/Purpose: This study was designed to determine the volume, postoperative surgical outcomes and, if possible, the relationship between outcome and institutional / surgeon volume in neonates undergoing repair of esophageal atresia with tracheoesophageal fistula (EA-TEF) over the last 20 years in Ontario.
Methods: Using administrative databases, a population based cohort study of patients undergoing EA-TEF repair in Ontario between 1993 and 2012 was conducted.
Results: 465 patients with the diagnosis of EA-TEF met inclusion criteria. The mean number of EA-TEF repairs per year per was 5.8. There was a significant difference in hospital annual volume between institutions (range 12.3–3.35: $p < 0.05$). The average number of cases/surgeon for the last 10 study years ranged between 0.5 and 2 cases/year. Primary outcome revealed that repair of recurrent fistula or intestinal interposition was 5.3%, with no reportable difference between institutions. Secondary outcomes revealed that 45.6% underwent dilatation for esophageal strictures, and 19.8% underwent some type of drainage procedure of the chest. These rates were not significantly different between institutions.
Conclusion: This study provides insight into the outcomes following EA-TEF repair in Ontario and the difficulty in determining surgeon or institution volume outcome relationships, as both primary and secondary outcome event rates are very low.
Level of Evidence: 2.

1. Background

Esophageal atresia with tracheoesophageal fistula (EA-TEF) is a rare congenital anomaly. The anomaly results from an insult occurring within the fourth week of gestation, during which time separation of the trachea and esophagus usually occurs by folding of the primitive foregut [1,2]. Most cases occur sporadically without evidence of either hereditary or specific environmental teratogenic causes [3–5]. The incidence of EA-TEF varies between 2.55 and 2.82 per 10,000 births [3–5].

Surgical intervention is required to establish esophageal continuity and ligate the fistula and was first reported in 1943 [6]. The pitfalls of the operation and the incidence of complications, both short term and long term, have been reported by many pediatric surgeons around the world. However, most of the studies are retrospective single center chart reviews [7–12].

Variations in surgical care and outcomes are very common in both pediatric and adult surgical practice. The relationship between the attending surgeons' volume of surgical procedures and surgical outcome has been studied extensively in the adult literature [13]. However, there are only a few studies in the pediatric population that have examined the volume outcome relationship, with one in the area of pediatric cardiac surgery [14] and another investigating outcomes following repair of congenital diaphragmatic hernia [15,16]. Others have compared surgical outcomes between procedures carried out by specialty trained pediatric surgeons and non-specialty-trained surgeons [17–19].

The purpose of this study was first to determine the number of EA-TEF repairs in Ontario over the last 20 years, and second to determine

Abbreviations: EA-TEF, Esophageal atresia with tracheoesophageal fistula; ICES, Institute for Clinical Evaluative Sciences; CIHI, Canadian Institute for Health Information's; DAD, Discharge Abstract Data base; SDS, Same Day Surgery Database; OHIP, Ontario Health Insurance Plan; RPDB, Registered Persons Database; GERD, Gastroesophageal reflux; UGI, Upper Gastrointestinal Study.

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https://doi.org/10.1016/j.jpedsurg.2018.02.019
0022-3468/

the frequency and volume outcome relationship of a number of postoperative complications and interventions in children undergoing EA-TEF repair, utilizing data from well-established province-wide healthcare databases held at the Institute for Clinical Evaluative Sciences (ICES).

2. Methods

2.1. Study design

This population based, retrospective cohort study included all pediatric patients undergoing EA-TEF repair at 4 pediatric centers in Ontario (Hospital for Sick Children in Toronto, Children's Hospital of Eastern Ontario in Ottawa, McMaster Children's Hospital in Hamilton, Children's Hospital at London Health Sciences Centre in London) between April 1st 1993 and March 31st 2013. Residents of Ontario have universal access to hospital care and these encounters are recorded in large population-based health care databases, dating back to 1988. Administrative datasets were linked using unique, encoded identifies and analyzed at ICES Western. Ethics approval was granted through the Sunnybrook Health Sciences Centre Research Ethics Board.

2.2. Data sources

Diagnostic and procedural information for all hospitalizations and one-day procedures was obtained from the Canadian Institute for Health Information's (CIHI) discharge abstract (DAD) and same day surgery (SDS) databases, whereas physician billing records were obtained from the Ontario Health Insurance Plan (OHIP). Patient characteristics were obtained from the Registered Persons Database (RPDB). Cases were identified using codes of EA-TEF from International Classification of Diseases, Ninth and Tenth Revision and procedure codes for EA-TEF surgical repair procedures using the Canadian Classification of Health Interventions.

2.3. Cohort build

All patients undergoing a primary EA-TEF repair between April 1st, 1993 and March 31st, 2013 at one of the 4 pediatric centers listed above were identified. All pediatric patients' resident in Ontario aged 180 days or younger who were Ontario residents when the index (initial EA-TEF repair) procedure was performed were included in the study.

Patients were excluded if they were not registered in ICES, if sex or date of birth was missing, if death occurred prior to the index event, or if there was no diagnosis of EA-TEF associated with the hospital discharge date within 60-days prior to the index procedure (including the procedure date) in DAD or SDS. Patients were also excluded if the encounter was associated with evidence of other esophageal injury or malignancy within 60-days before the index date (including the index date) in DAD or SDS.

2.4. Data collection

Baseline variables such as age of patient at time of primary procedure, location of surgery and sex of patient were collected. The number of surgeons per institution for period of study was obtained by personal communication (Dr S Jones) with each center.

2.4.1. Outcomes

The primary outcome was a composite of repeat EA-TEF repair and surgery for definitive reconstruction of the esophagus (gastric pull-up or intestinal pull-up) within two-years of the initial EA-TEF repair. Secondary outcomes included other surgical procedures of the bronchus, trachea and esophagus, insertion of a gastrostomy tube, the procedure of dilatation of esophageal stricture, assessment of esophageal patency /motility/gastroesophageal reflux (GERD) by radiological contrast study (upper gastrointestinal study (UGI)) and "other" surgical interventions that occurred and were thought to be related to EA-TEF repair, all within two-years of the initial procedure.

2.5. Analysis

Data are reported for the complete 20-year period, stratified by 5-year periods and by center. Owing to ICES privacy requirements, institution names have been anonymized (as A, B, C, and D) and data representing fewer than 6 individuals (including percentages) have been suppressed throughout as not reportable (NR). Between center differences in baseline characteristics and outcomes were evaluated using the chi-square test for association, except continuous baseline variables which were assessed using ANOVA or Kruskal Wallis, as appropriate. For all analyses, reported p-values are from two-sided tests where a value of <0.05 was considered statistically significant. All analyses were performed using SAS EG version 7.1 (SAS Institute, Cary, NC, USA).

3. Results

3.1. Patient and center characteristics

A total of 465 patients from the 4 centers met the criteria for inclusion in the primary cohort (both diagnosis of EA-TEF and procedure for EA-TEF repair: Table 1). As expected, all primary procedures were completed during an inpatient admission. Of the primary cohort 41.5% of patients were females and 58.5% were males. The median age at the time of the index surgery was 2 days (quartile range 1–4); this was not significantly different between centers.

The average number of primary EA-TEF repairs over the study period was 23/year. The number of EA-TEF repairs for the eras: 1993–97, 1998–2002, 2003–2007 and 2008–2013 was 126, 122, 113 and 104 respectively. There was a statistically significant difference in patient volume per institution for the 20-year period of study (Table 2). The average number of primary EA-TEF repairs per surgeon was calculated (number of EA-TEF repairs per institution divided by the average number of surgeons per 5-year block) to be approximately 2/year in 1993–1997; this number decreased over time to approximately 1 case per surgeon per year by era 2008–2012 (Fig. 1).

3.2. Outcomes

3.2.1. Primary outcomes

Five percent ($n = 25/465$) of the primary cohort required reoperation, 16 for recurrent trachea–esophageal fistula and 10 for esophageal replacement with an intestinal interposition.

3.2.2. Secondary outcomes

An upper GI study was performed in 27.1% patients and was not significantly different between institutions. Dilatation for esophageal stricture was reported in 45.6% of TEF patients (range 41.9%–53.4% between institutions) and a gastrostomy tube was performed on 144 (31%) of

Table 1
EA-TEF cohort build.

Step	Number excluded	Number included
Original cohort	Before Exclusion	825
Missing sex or age	9	816
Death prior to index date	0	816
Age ≥ 180 days	322	494
Non-Ontario resident	NR (included in step below)	suppressed
No diagnosis of EA-TEF 60 days prior to index date	NR (included in step below)	suppressed
Procedure not performed at pediatric teaching	NR (included in step below)	suppressed
Evidence of other esophageal injury	29	465