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Challenging surgical dogma in the management of proximal esophageal atresia with distal tracheoesophageal fistula: Outcomes from the Midwest Pediatric Surgery Consortium

Dave R. Lal [a,*], Samir K. Gadepalli [b], Cynthia D. Downard [c], Daniel J. Ostlie [d], Peter C. Minneci [e], Ruth M. Swedler [a], Thomas H. Chelius [f], Laura Cassidy [a,f], Cooper T. Rapp [a], Deborah Billmire [g], Steven Bruch [b], R. Carland Burns [g], Katherine J. Deans [e], Mary E. Fallat [c], Jason D. Fraser [h], Julia Grabowski [i], Ferdynand Hebel [i], Michael A. Helmrath [j], Ronald B. Hirschl [b], Rashmi Kabre [i], Jonathan Kohler [d], Matthew P. Landman [g], Charles M. Leys [d], Grace Z. Mak [k], Jessica Raque [c], Beth Rymeski [j], Jacqueline M. Saito [l], Shawn D. St. Peter [h], Daniel von Allmen [j], Brad W. Warner [l], Thomas T. Sato [a], on behalf of the Midwest Pediatric Surgery Consortium

[a] Division of Pediatric Surgery, Department of Surgery, Medical College of Wisconsin, Milwaukee, WI
[b] Section of Pediatric Surgery, Department of Surgery, University of Michigan, Ann Arbor, MI
[c] Division of Pediatric Surgery, Hiram C. Polk, Jr., M.D. Department of Surgery, University of Louisville, Louisville, KY
[d] Division of Pediatric Surgery, Department of Surgery, University of Wisconsin, Madison, WI
[e] Center for Surgical Outcomes Research, The Research Institute at Nationwide Children's Hospital, Department of Surgery, University of Ohio, Columbus, OH
[f] Division of Epidemiology, Institute for Health and Society, Medical College of Wisconsin, Milwaukee, WI
[g] Division of Pediatric Surgery, Department of Surgery, Indiana University School of Medicine, Indianapolis, IN
[h] Division of Pediatric Surgery, Department of Surgery, Children's Mercy Hospital, Kansas City, MO
[i] Division of Pediatric Surgery, Department of Surgery, Ann & Robert H. Lurie Children's Hospital of Chicago, Northwestern University Feinberg School of Medicine, Chicago, IL
[j] Division of Pediatric Surgery, Cincinnati Children's Hospital Medical Center, Cincinnati, OH
[k] Section of Pediatric Surgery, Department of Surgery, The University of Chicago Medicine and Biologic Sciences, Chicago, IL
[l] Division of Pediatric Surgery, Department of Surgery, Washington University School of Medicine, St. Louis, MO

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ABSTRACT

Purpose: Perioperative management of infants with esophageal atresia and tracheoesophageal fistula (EA/TEF) is frequently based on surgeon experience and dogma rather than evidence-based guidelines. This study examines whether commonly perceived important aspects of practice affect outcome in a contemporary multi-institutional cohort of patients undergoing primary repair for the most common type of esophageal atresia anomaly, proximal EA with distal TEF.

Methods: The Midwest Pediatric Surgery Consortium conducted a multicenter, retrospective study examining selected outcomes on infants diagnosed with proximal EA with distal TEF who underwent primary repair over a 5-year period (2009–2014), with a minimum 1-year follow up, across 11 centers.

Results: 292 patients with proximal EA and distal TEF who underwent primary repair were reviewed. The overall mortality was 6% and was significantly associated with the presence of congenital heart disease (OR 4.82, $p = 0.005$). Postoperative complications occurred in 181 (62%) infants, including: anastomotic stricture requiring intervention ($n = 127$; 43%); anastomotic leak ($n = 54$; 18%); recurrent fistula ($n = 15$; 5%); vocal cord paralysis/paresis ($n = 14$; 5%); and esophageal dehiscence ($n = 5$; 2%). Placement of a transanastomotic tube was associated with an increase in esophageal stricture formation (OR 2.2, $p = 0.01$). Acid suppression was not associated with altered rates of stricture, leak or pneumonia (all $p > 0.1$). Placement of interposing prosthetic material between the esophageal and tracheal suture lines was associated with an increased leak rate (OR 4.7, $p < 0.001$), but no difference in the incidence of recurrent fistula ($p = 0.3$). Empiric postoperative antibiotics for >24 h were used in 193 patients (66%) with no difference in rates of infection, shock or death when compared to antibiotic use ≤24 h (all $p > 0.3$). Hospital volume was not associated with postoperative complication rates ($p > 0.08$). Routine postoperative esophagram obtained on day 5 resulted in no delayed/missed anastomotic leaks or a difference in anastomotic leak rate as compared to esophagrams obtained on day 7.

Conclusion: Morbidity after primary repair of proximal EA and distal TEF patients is substantial, and many common practices do not appear to reduce complications. Specifically, this large retrospective series does not support the use of prophylactic antibiotics beyond 24 h and empiric acid suppression may not prevent complications. Use of a transanastomotic tube was associated with higher rates of stricture, and interposition of prosthetic material

* Corresponding author.
E-mail address: dlal@chw.org (D.R. Lal).

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was associated with higher leak rates. Routine postoperative esophagram can be safely obtained on day 5 resulting in earlier initiation of oral feeds.
Study type: Treatment study.
Level of evidence: III.

Repair of esophageal atresia (EA) with distal tracheoesophageal fistula (TEF) is a quintessential operation that defines pediatric surgery. Although mortality associated with EA repair has markedly decreased since the operation was first described, an improving trend in postoperative complications has been less clear. Contemporary perioperative management of infants with EA is generally based upon surgeon experience rather than evidence-based guidelines. As a result, there is wide variation in management among surgeons.

This study aims to define variation in perioperative management and complications in infants with the most common configuration of esophageal atresia — proximal EA with distal TEF (Gross type C). By identifying associations between perioperative management variation and complications, we seek to further understand the benefits, if any, of generally accepted surgical practice, including duration of antimicrobial prophylaxis, placement of transanastomotic tubes, use of acid suppression, and timing of postoperative esophagram. Finally, we hypothesize that there are management variables amenable to modification which result in reduced complications and improved outcomes.

1. Methods

1.1. Patients and study design

Following individual institutional review board approval, a retrospective cohort of infants diagnosed with esophageal atresia and/or tracheoesophageal fistula between 2009 and 2014 was identified across 11 participating children's hospitals of the Midwest Pediatric Surgery Consortium (see Acknowledgments for list of centers). We have previously reported on this entire cohort with proximal reported on this entire cohort of 396 infants [1,2]. This study examines a subset of that cohort with proximal esophageal atresia and distal tracheoesophageal fistula who underwent primary esophageal repair. All infants with proximal esophageal atresia with distal tracheal esophageal fistula (EA/TEF) (ICD-9750.3) diagnosed within 30 days of life who underwent primary repair of the esophageal atresia within the first six months of life from January 1, 2009 to January 1, 2014 were included. Patients were identified from administrative hospital databases as well as practice databases. A minimum of 1-year follow up was obtained excluding those who died before the age of 1 year.

Study data were collected and managed using REDCap (Research Electronic Data Capture) software hosted at The Medical College of Wisconsin [3]. All study data entered were validated both centrally and at each individual institution for completeness of data entry and accuracy. Missing data were reobtained to the degree possible, and outliers were reconfirmed to be accurate.

1.2. Methods

Consensus in identifying and defining relevant data elements for collection and analysis across the 11 institutions was established prior to data collection. Perioperative data collection included demographics, prenatal information when available, associated congenital anomalies, diagnostic imaging, physiologic status including American Society of Anesthesiology (ASA) Physical Status Classification at the time of operation, operative approach and findings, postoperative morbidity and mortality, and need for further intervention(s) up to one year following repair. Race/ethnicity was self-reported by patients during hospital registration. Congenital heart disease was defined as clinically identified cardiac defects other than patent ductus arteriosus and patent foramen ovale. Long gap esophageal atresia was defined as a distance of three or more vertebral bodies between the proximal and distal esophageal segments determined on preoperative chest x-ray/esophageal pouch contrast studies or at the time of surgical exploration. Esophageal anastomotic stricture was defined as the need for dilatation of any type within one year of establishing esophageal continuity. Prenatal diagnosis or suspicion of EA/TEF was determined by reviewing the prenatal ultrasound reports for mention of polyhydramnios and/or absent/small stomach with mention of a possible diagnosis of EA within the report.

1.3. Statistical analysis

Statistical analysis was performed using SAS version 9.4 (SAS Institute Inc., Cary, NC, USA) and STATA (StataCorp. 2011. Stata Statistical Software: Release 12. College Station, TX: StataCorp LP). Missing data were recollected to ensure accuracy when possible; patients with missing data were excluded from the analysis. Continuous data were expressed as medians and interquartile ranges, and discrete variables were expressed as percentages. For discrete variable comparisons, Pearson's Chi-Square Test was used. Fisher's Exact Test was used for associations of two binary variables which had small cell sizes. The Wilcoxon Rank-Sum test and Kruskal–Wallis test were used to compare medians. A logistic regression was performed for outcomes, using variables defined by consensus for entry into the model or a p-value <0.2 on univariate analysis. A priori specified variables based on consensus to be included in all models were institution site, the presence of congenital heart disease and the presence of long gap esophageal atresia. However, institution site and use of a transanastomotic tube were collinear in the model for stricture formation. Since transanastomotic tube explained more of the variability in the model, it was retained in the model and site was removed. To further explore the relationship between use of a transanastomotic tube and stricture formation, a Cochran–Armitage test for trend was performed based on the reported frequency of transanastomotic tube use at each center categorized as low (used in $<20\%$ of cases), moderate (used in 20% to 79% of cases), and high (used in $\geq 80\%$ of cases). A p-value less than 0.05 was considered statistically significant.

2. Results

2.1. Study cohort and demographics

Over the five-year study period, 292 infants with proximal esophageal atresia and distal tracheoesophageal fistula (EA/TEF) underwent primary esophageal repair among the 11 children's hospitals. On average, 5 cases were identified per institution per year of the study. Prenatal diagnosis of the EA/TEF was diagnosed or suspected in 25 patients (8.6%).

Of the 292 infants, 176 (60%) were male; 227 (78%) were Caucasian, 21 (7%) African American, 18 (6%) Hispanic, 7 (2%) Asian, 7 (2%) Multiracial, and the remainder were other or not reported. One hundred fifty-three patients (52%) had private insurance, 126 (43%) public insurance, 13 (4%) self-pay or unknown. The median gestational age at birth was

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