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Foker process for the correction of long gap esophageal atresia: Primary treatment versus secondary treatment after prior esophageal surgery

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ABSTRACT

Purpose: The Foker process (FP) uses tension-induced growth for primary esophageal reconstruction in patients with long gap esophageal atresia (LGEA). It has been less well described in LGEA patients who have undergone prior esophageal reconstruction attempts.
Methods: All cases of LGEA treated at our institution from January 2005 to April 2014 were retrospectively reviewed. Patients who initially had esophageal surgery elsewhere were considered secondary FP cases. Demographics, esophageal evaluations, and complications were collected. Median time to esophageal anastomosis and full oral nutrition was estimated using the Kaplan–Meier method. Multivariate Cox-proportional hazards regression identified potential risk factors.
Results: Fifty-two patients were identified, including 27 primary versus 25 secondary FP patients. Median time to anastomosis was 14 days for primary and 35 days for secondary cases ($p < 0.001$). Secondary cases ($p = 0.013$) and number of thoracotomies ($p < 0.001$) were identified as significant predictors for achieving anastomosis and the development of a leak. Predictors of progression to full oral feeding were primary FP cases (O.R. = 17.0, 95% CI: 2.8–102, $p < 0.001$) and patients with longer follow-up (O.R. = 1.06/month, 95% CI: 1.01–1.11, $p = 0.005$).
Conclusions: The FP has been successful in repairing infants with primary LGEA, but the secondary LGEA patients proved to be more challenging to achieve a primary esophageal anastomosis. Early referral to a multidisciplinary esophageal center and a flexible approach to establish continuity in secondary patients is recommended. Given their complexity, larger studies are needed to evaluate long-term outcomes and discern optimal strategies for reconstruction.

The difficulties in treating long gap esophageal atresia (LGEA) patients are well known, as are the controversies surrounding the operative repair. LGEA has not been defined precisely but includes any patient with esophageal atresia (EA) that cannot undergo an initial primary repair. Although, the definition of what constitutes LGEA has not been agreed upon, the overall goal is universal; to achieve a functional esophagus that allows for normal eating with lifelong durability. More recently in LGEA patients, axial tension on the proximal and distal esophageal segments has been shown to reliably induce sufficient esophageal growth to allow for a primary esophageal repair [1,2]. Initially described in 1997, the Foker process (FP) can be technically demanding; however, particularly when the atretic lower segments are very small [2].

One criticism of the FP is the relative rarity of LGEA cases overall, surgeon comfort-level and expertise, and, consequently, few centers have the patient volume required to construct, refine, and maintain the necessary skills in a multidisciplinary team dedicated to the treatment of these infants and young children [3]. Our own institution started utilizing the FP primarily for all LGEA patients in 2005; even less has been described in LGEA patients who have undergone prior reconstruction attempts and their outcomes versus primary repairs. Inherently, there is an added level of difficultly from prior reconstruction attempts, as well as unforeseen intricacies related to poor nutrition, access-related issues, and developmental concerns. Therefore, the purpose of this study is to 1) compare these two distinct cohorts to ascertain and evaluate potential differences in short-term outcomes, as well as complications; and, 2) suggest the best strategies for approach to both primary and secondary LGEA patients.

1. Methods

1.1. Basic demographics

Following institution review board approval (IRB Protocol M10-10-052), we retrospectively reviewed all cases of LGEA who were treated

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from January 2005 to April 2014 at Boston Children's Hospital (BCH). Primary LGEA patients were those patients who did not undergo a previous operation or whose previous operations were limited to a gastrostomy placement. Those patients who had esophageal surgery elsewhere were considered secondary FP cases. Previous operations included thoracotomies with repair of proximal and distal tracheoesophageal fistulas (TEF), primary repair of esophageal atresia, attempted repair of esophageal atresia by Foker process (FP) and esophageal replacements. Previous operations also included those related to secondary complications including mediastinitis, chylothoraces, empyemas, recalcitrant strictures, and dilation-related perforations.

Patient-data collected included: basic demographics, associated anomalies, gap length, time to complete Foker process (FP), intensive care (ICU) data, number of thoracotomies, stricture treatment and dilations, complications, length of follow-up and patient outcome. Patient outcomes were further subdivided into the following: attainment of a functional native esophagus, or whether an interposition was required; and, eating by mouth solely versus supplementation and/or primarily enteral feeds. Complications recorded included symptomatic venous thromboembolic events (VTE) and fractures. Routine screening was not performed for these events. Mortality was also recorded.

1.2. Operative technique

The Foker process was conducted by the surgeons on the esophageal atresia multidisciplinary team on both primary and secondary cases through a 3–4 centimeter posterior thoracotomy incision [2]. Both upper and lower esophageal segments were identified and mobilized within the right pleural space. The 3rd and 7th intercostal spaces were opened utilizing the same skin incision in the cases of longer gaps. Pledgeted traction sutures of 5-0 or 6-0 Prolene® were placed in the upper and lower esophageal segments for external traction. Sutures were placed through the muscular and submucosal layers. The esophageal segments were enclosed in silastic sheeting. Tension was increased daily at the bedside by placing segments of feeding tubes under the sutures. Movement of clips placed on the esophageal segments was monitored by serial radiographs. Weekly contrast studies were performed to confirm that the lumen was lengthening along with the esophageal wall and to identify potential esophageal leaks. Repeat thoracotomies were done when replacement and reconfiguration of the sutures were needed to reestablish tension, as well as when an esophageal anastomosis or interposition was performed.

1.3. Statistical analysis

Univariate analysis was used to compare demographic and clinical data as well as patient outcomes including achievement of esophageal anastomosis, venous thrombotic events (VTE), fractures, full oral nutrition, reoperation rates, and mortality. Birth weight, gap length, intensive care (ICU) and hospital stay, ventilation days, number of dilations and thoracotomies were compared between primary and secondary FP cases using the Mann–Whitney *U*-test with data summarized using the median and range. Simple proportions were compared by Fisher's exact test for binomial data. Kaplan–Meier time-to-event analysis was performed to compare time to achievement of esophageal anastomosis and freedom from fractures between primary and secondary FP cases with the log-rank test to compare the curves and Greenwood's formula to calculate 95% confidence intervals [4].

Multivariable logistic regression was applied to identify independent predictors of esophageal anastomosis and leaks in order to control for possible confounding with odds ratios (OR) and 95% CIs for significant predictors [5]. Statistical analysis was conducted using IBM SPSS Statistics (version 21.0, IBM, Armonk, NY). Two-tailed values of $p < 0.05$ were considered statistically significant. Power analysis indicated that a minimum of 25 primary and 25 secondary FP cases would provide 80% power to detect 30–40% differences with respect to patient outcomes including anastomosis, leaks, VTEs and fractures using Fisher's exact test (version 7.0, nQuery Advisor, Statistical Solutions, Saugus, MA).

2. Results

Fifty-two patients were analyzed during this study period. Twenty-seven were classified as primary FP patients and twenty-five were classified as secondary FP patients. These latter patients presented from 5 US states and 3 other countries. Reported birth weight (BW), estimated gestational age (EGA), and estimated gap length were similar between the two cohorts at baseline. There were also no significant differences in gender, cardiac defects, and baseline VACTERL phenotypes. Weight for age Z scores, a marker for nutritional status, was not significantly different at hospital admission; median Z-scores of −1.26 for primary FP patients versus −1.18 for secondary FP patients respectively. The median weight at the FP was 5 kg (4.1–7.9 kg). There was not a significant difference between the two cohorts. The median age at time of Foker process was 4 months (range: 2–7 months) (Table 1)

Eighteen (67%) of the primary FP presented as “pure”, or isolated LGEA patients. The remaining 9 (33%) patients had a proximal fistula. Of the 25 secondary FP patients, 13 had a failed FP process, 10 patients had a failed type C-EA primary repair and 2 had a failed colonic interpositions. Three primary FP patients had Trisomy-21 (Down's syndrome) versus 4 patients within the secondary FP cohort. Within the secondary FP patients, 8 had a cervical esophagostomy (5 right-sided and 3 left-sided esophagostomies). Three patients presented with a tracheostomy and 24% (n = 6) of the secondary FP patients had either single or bilateral vocal cord paresis/paralysis. Median attempts at FP repair prior to hospitalization at our institution were 2 (range: 1–4 attempts) and the complications included 1 esophageal stent erosion and 2 empyemas.

Median time (days) to initiating the FP after admission was different between cohorts; repair of the primary patients was begun after 24 days (range: 1–144 days) and by 8 days (range: 1–361 days) for secondary patients. The number of thoracotomies differed at baseline between the two groups ($p < 0.001$) with the secondary FP cases requiring more thoracotomies during their hospital stay. Median time from

Table 1

Characteristics of primary and secondary LGEA patients

Characteristic	Primary FP cases (n = 27)	Secondary FP cases (n = 25)	P value
Birth weight, kg	2.3 (0.8–4.6)	2.9 (1.5–3.7)	0.7
Gestational age, weeks	37 (25–39)	36 (29–39)	0.4
Estimated gap length, cm	4.5 (2.9–6.0)	5.0 (1.6–9.0)	0.2
Male gender	17 (63%)	12 (48%)	0.4
Cardiac defects	11 (41%)	9 (36%)	0.7
VACTERL	10 (37%)	9 (36%)	1.0
Hospital stay, days	108 (22–269)	134 (64–685)	0.03*
ICU stay, days	70 (22–217)	110 (35–685)	0.04*
Paralytics, days	17 (0–64)	44 (0–133)	<0.001*
Mechanical ventilation, days	24 (15–173)	46 (9–236)	0.005*
VTE	3 (11%)	12 (48%)	0.005*
Fractures	5 (19%)	15 (60%)	0.004*
# of thoracotomies	2 (2–10)	5 (2–15)	<0.001*
# of dilations in hospital**	3 (0–18)	5 (0–20)	0.6
Intact esophagus	26 (96%)	17 (68%)	0.01*
Full oral nutrition	17 (63%)	2 (9%)	<0.001*
Mortality	0 (0%)	2 (8%)†	0.2

Continuous data are expressed as median (range). ICU = intensive care unit; LGEA = long gap esophageal atresia, VTE = venous thromboembolism.

* Statistically significant.

** Number of dilations during the primary hospital stay for original Foker process.

† No patients died in hospital, however two patients with complex anatomy with failed attempts at repair died after discharge (one had multiple additional complex medical problems).