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Primary Posterior Tracheopexy at Time of Esophageal Atresia Repair Significantly Reduces Respiratory Morbidity

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ABSTRACT

Purpose: Esophageal atresia with tracheoesophageal fistula (EA/TEF) is often associated with tracheobronchomalacia (TBM), which contributes to respiratory morbidity. Posterior tracheopexy (PT) is an established technique to treat TBM that develops after EA/TEF repair. This study evaluates the impact of primary PT at the time of initial EA/TEF repair.

Methods: Review of all newborn primary EA/TEF repairs (2016–2021) at two institutions. Long-gap EA and reoperative cases were excluded. Based on surgeon preference and preoperative bronchoscopy, neonates underwent primary PT (EA + PT Group) or not (EA Group). Perioperative, respiratory and nutritional outcomes within the first year of life were evaluated.

Results: Among 63 neonates, 21 (33%) underwent PT during EA/TEF repair. Groups were similar in terms of demographics, approach, and complications. Neonates in the EA + PT Group were significantly less likely to have respiratory infections requiring hospitalization within the first year of life (0% vs 26%, $p = 0.01$) or blue spells (0% vs 19%, $p = 0.04$). Also, they demonstrated improved weight-for-age z scores at 12 months of age (0.24 vs −1.02, $p < 0.001$). Of the infants who did not undergo primary PT, 10 (24%) developed severe TBM symptoms and underwent tracheopexy during the first year of life, whereas no infant in the EA + PT Group needed additional airway surgery ($p = 0.01$).

Conclusion: Incorporation of posterior tracheopexy during newborn EA/TEF repair is associated with significantly reduced respiratory morbidity within the first year of life.

Level of evidence: Level III.

1. Introduction

While survival for infants with esophageal atresia with tracheoesophageal fistula (EA/TEF) has improved dramatically, morbidity remains high. Tracheobronchomalacia (TBM) represents a significant source of morbidity for this population, with a reported prevalence of up to 87% [1,2]. TBM is defined as excessive dynamic collapse of the trachea and bronchi, particularly on exhalation, that results in impaired air flow and mucociliary clearance. Unfortunately, there is the general misperception, despite lack of data to support the notion, that infants with congenital TBM will outgrow their symptoms over time. Though this may be possible for some, we lack the ability to predict who will and who will not improve, and the burden on the child and family while they "wait it out" can be significant. In fact, some infants with severe TBM manifestations, such as failure to extubate, failure to wean respiratory support, or who have brief resolved unexplained events (BRUEs or blue spells), are often treated with a tracheostomy [3]. Others endure chronic barky cough, noisy breathing, recurrent respiratory infections, poor oral feeding, growth impairment, chronic respiratory compromise, and more.

Our groups have approached TBM in a systematic and multidisciplinary approach that involves early identification, accurate classification and tailored treatment. Though some respond to medical management, surgical intervention is offered to those with

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moderate to severe symptoms. Historically, surgery for TBM involved anterior aortopexy [4]. However, results were mixed, likely because it did not directly address the posterior membranous tracheal intrusion, which is the hallmark of TBM seen in children with a history of EA/TEF. More recently, posterior tracheopexy (PT) has emerged as an established operative intervention for TBM [5]. Posterior tracheopexy was initially described at our institution in the management of recurrent TEF, eliminating the risk of re-recurrence by effectively separating the tracheal and esophageal suture lines [6,7]. On subsequent bronchoscopic evaluations, children who had undergone PT were noted to have marked improvement in posterior intrusion type tracheomalacia. We thus began using PT to surgically treat TBM, particularly in children who had previously undergone EA/TEF repair. Our experience with this technique has shown promising short- and long-term results [8–11]. To date, however, few studies have evaluated the role for PT in the neonate at the time of index EA/TEF repair [12,13]. The objective of this study was to investigate the impact of incorporating posterior tracheopexy at the time of initial newborn EA/TEF repair on respiratory and perioperative outcomes, particularly within the first year of life. We hypothesized that primary tracheopexy at the time of EA/TEF repair would be associated with improved respiratory outcomes.

2. Methods

2.1. Study design

With Institutional Review Board (IRB) approval, we conducted a retrospective review of all consecutive newborns with Gross Type C or D EA/TEF who were surgically treated with primary EA repair between the years 2016 and 2021 at Boston Children's Hospital (BCH, Boston, MA) or between the years of 2019 and 2021 at Johns Hopkins All Children's Hospital (JHACH, St. Petersburg, Florida). Neonates with EA/TEF requiring a staged approach or traction-induced esophageal lengthening (Foker process) and/or reoperation were excluded, in order to truly focus on those in whom there had been no time period to assess for TBM symptoms. Based on surgeon preference and findings on preoperative bronchoscopy, neonates underwent primary posterior tracheopexy during the initial EA/TEF repair (EA + PT Group) or not (EA Group).

2.2. Surgical procedure

Newborns underwent a diagnostic laryngoscopy and rigid dynamic tracheobronchoscopy immediately prior to EA/TEF repair, if possible, to evaluate for laryngeal cleft, aspirate secretions from within the tracheobronchial tree, assess for the presence of a concomitant proximal/cervical TEF, evaluate the presence and degree of tracheobronchomalacia, and attempt Fogarty balloon occlusion of the distal TEF. TBM was classified as mild, moderate, or severe based on the percent of intrusion of the posterior membrane into the airway lumen using our previously described classification system (Fig. 1) [5]. Newborns were subsequently intubated with a suitable size endotracheal tube over a rigid bronchoscope to assure appropriate placement of the endotracheal tube (ETT) in relation to the TEF. Newborns then underwent either open or minimally invasive repair at the discretion of the operating surgeon. After the fistula was divided, the airway was repaired with absorbable monofilament suture in interrupted fashion. Based on surgeon preference and initial bronchoscopy, newborns either underwent primary PT or not.

Our surgical technique for PT has been previously described [5]. In brief, the posterior membrane of the airway is exposed, the process of which requires adequate circumferential dissection of the esophagus into the thoracic inlet while being mindful of the recurrent laryngeal nerves. After this, the anterior longitudinal ligament of the spine is exposed, taking caution to minimize risk of injury to the thoracic duct and sympathetic plexus. Autologous tissue (pleura or azygos vein) -pledgeted non-absorbable (polypropylene) sutures are then passed into (but not through) the posterior membrane of the trachea in horizontal mattress fashion using flexible bronchoscopic guidance, securing the posterior membrane of the trachea to the anterior longitudinal ligament of the spine. For newborns included in this study, tracheopexy was performed in the area of the TEF repair tracheal suture line and in any additional locations warranted based on the initial bronchoscopy. After completion of the tracheopexy, the esophageal atresia was repaired with a primary esophago-esophageal anastomosis per surgeon preference, keeping the esophagus to the right side of the tracheopexy.

2.3. Outcome measures

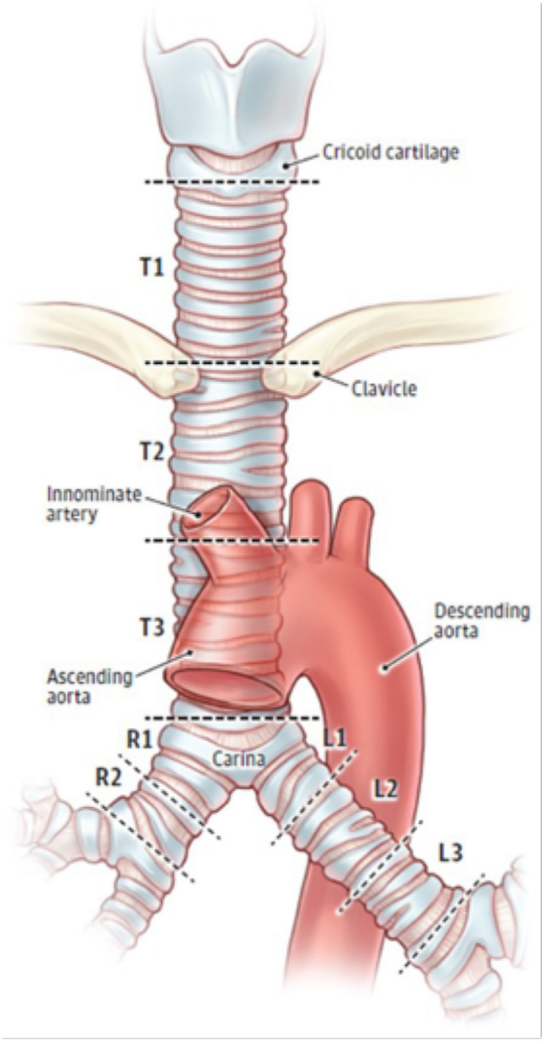
Demographic, perioperative, and follow-up data were abstracted from the electronic medical record. Our primary outcome of interest pertained to respiratory morbidity. This included post-operative time to extubation, need for reintubation, need for positive pressure ventilation (PPV) within the first week post-operatively, respiratory infections within the index hospitalization, and need for endoscopic airway evaluation within the index hospitalization. Beyond this initial admission, additional respiratory outcomes assessed included need for airway evaluations or bronchoscopies, respiratory infections with or without need for hospitalization, blue spells or BRUEs/ALTEs, aspiration events, oxygen dependence, barky cough, noisy breathing, stridor, wheezing, and need for delayed tracheopexy within the first year of life or at any point during the extended follow-up period. Additional outcomes of interest included length of hospital stay, length of stay in the Intensive Care Unit (ICU), operative time, incidence of various post-operative complications such as esophageal leak, esophageal stricture, chyle leak, subglottic stenosis, vocal fold movement impairment, feeding tube dependency at various points within the first post-operative year, and weight-for-age Z-scores at 12 months post-operatively. All post-operative esophagrams and any subsequent cross-sectional imaging were also reviewed to assess the location of the esophagus in relation to the midline. All post-operative bronchoscopies were reviewed to assess for findings, such as location and degree of tracheomalacia, presence of tracheal diverticulum, recurrent, acquired or missed TEF, among others.

2.4. Statistical analysis

Descriptive statistics were calculated and reported. Continuous variables were presented as median with the interquartile range; categorical data were presented as frequencies with their associated percentages. Univariate associations were compared with Wilcoxon rank-sum test for continuous variables and with Chi-square test for categorical variables. P-values less than 0.05 were considered statistically significant. Analyses were conducted using JMP v15 (SAS, Cary, NC).

3. Results

A total of 63 consecutive newborns underwent EA/TEF repair for Gross types C or D anomalies between January 1, 2016, and December 31, 2021, at BCH (n = 49) and between January 1, 2019, and December 31, 2021, at JHACH (n = 14). Of these 63 neonates, 21 (33%) underwent primary posterior tracheopexy at the time of initial EA/TEF repair (EA + PT Group). The remaining 42 did not



	FOE awake	Shallow breathing	Cough valsalva	Sedated distended pressure?	Contrast study
Tonsils					
Epiglottis		NL			
AE folds		NL			
Arytenoids		NL			
Cords		POST SCAR			
L. cleft		REPAIRED			
Subglottis		10% ELLIP			
T1		"U" 10% POST	30%	FLAT	
T2		"BOW" 10% R	20%	FLAT	
T3		"BOW" 10% L	10%	FLAT	
Carina		"BOW"		FLAT	
L1		"U" 40% R	60% R	40% R	
L2		"U"	30% POST		
L3		"U"	10% POST		
R1		"U"	10%		
R2		"U"	10%		
BI		"U"	10%		
Tracheal diverticulum?		POSTERIOR CARINA		5mm DEEP	NO
TEF?					NO
Posterior artery?		NO		NO	
Secretions		THICK			
Other					
Notes			SCABBARD LMB		

Fig. 1. The trachea is divided into three regions. T1, referring to the upper third or the cervical trachea, spans from the cricoid cartilage to the clavicles. T2, referring to the middle third of the trachea, includes the region from the clavicles to the takeoff of the innominate artery (visualized bronchoscopically as an anterior tracheal impression). T3, referring to the lower third of the trachea, spans from the innominate artery takeoff to the carina. The distal airways are further subdivided as shown in the image above. Shape of cartilages is described and degree of anterior and posterior intrusion is assessed, both at rest and with dynamic maneuvers such as cough.

undergo prophylactic PT (EA Group). Aside from slightly greater birth weight in the EA + PT Group (median 2840g [IQR 2562–3190g] vs 2400 [2020–2980g], p = 0.02), there were no significant differences between the two groups in terms of gender, gestational age, prematurity status, twin gestation, perinatal APGAR scores, or associated anomalies (Table 1).

3.1. Clinical and surgical outcomes on initial admission

There was no significant difference in age at time of EA repair or need for pre-operative intubation between the two groups (Table 2). A total of 60 (95%) newborns underwent a bronchoscopy at the time of EA repair with no difference between groups with

Table 1
Study population characteristics.

	EA + PT Group (n = 21)	EA Group (n = 42)	p-value
Male, n (%)	13 (61.9%)	20 (47.6%)	0.28
Gestational age, median (wks, IQR)	39 (37.5–39)	37 (35–39)	0.08
Premature (<37 wks GA), n (%)	4 (19.0%)	16 (38.1%)	0.16
Birth weight, median (IQR), grams	2840 (2565–3190)	2400 (2020–2980)	0.02
Twin gestation	1 (4.7%)	4 (9.5%)	0.47
Associated anomalies			
Trisomy 21	0	1 (2.4%)	1.00
VACTERL association	8 (38.1%)	20 (47.6%)	0.47
Cardiac anomalies	6 (28.6%)	14 (33.3%)	0.70
Gastrointestinal anomalies	2 (9.5%)	5 (11.9%)	0.08
Type C EA/TEF, n (%)	21 (100%)	41 (97.6%)	0.82

Table 2
Surgical details.

	EA + PT Group (n = 21)	EA Group (n = 42)	p-value
Age at time of EA/TEF surgery, median (IQR), days	1 (1–2)	1 (1–2)	0.72
Pre-op intubation, n (%)	2 (9.5%)	10 (23.8%)	0.31
Pre-op bronchoscopy, n (%)	21 (100%)	39 (92.9%)	0.54
No TBM, n (%)	1 (4.8%)	14 (35.9%)	0.01
Mild TBM, n (%)	5 (23.8%)	20 (51.3%)	0.07
Moderate-Severe TBM, n (%)	13 (61.9%)	5 (12.8%)	<0.001
Location of TEF[a]			
T1, n (%)	1 (4.8%)	1 (2.6%)	1.00
T2, n (%)	9 (42.9%)	15 (38.5%)	0.69
T2/T3, n (%)	1 (4.8%)	2 (5.1%)	1.00
T3, n (%)	5 (23.8%)	7 (17.9%)	0.74
Carina, n (%)	5 (23.8%)	14 (35.9%)	0.39
Operative time, median (IQR), minutes	279 (240–351)	286 (217–324)	0.57
Right-sided approach, n (%)	20 (95.2%)	42 (100%)	0.33
Thoracoscopic approach, n (%)	2 (9.5%)	13 (31.0%)	0.07
Presence of aberrant right subclavian artery, n (%)	0 (0%)	2 (4.8%)	0.55
Subspecialized thoracic primary surgeon, n (%)	21 (100%)	19 (45.2%)	<0.001

[a] Location of TEF based on anatomic landmarks described in previously published work (Kamran et al. [24]).

respect to the location of the TEF (Table 2). Newborns in the EA + PT Group were more likely to have had moderate to severe tracheomalacia noted on initial dynamic bronchoscopy than those in the EA Group (n = 13, 61.9% vs n = 5, 12.8%, p < 0.001, Table 2). The majority of newborns underwent an open repair with a thoracotomy (n = 48, 76.2%) via the right chest (n = 62, 98%), with no difference between the two groups. Operative times were similar as well. All newborns underwent TEF repair and primary esophagoesophageal anastomosis.

Post-operatively, there were no significant differences between the two groups in terms of time to extubation, need for immediate reintubation (within 72 h post-extubation), need for positive pressure support within the first week post-extubation, incidence of respiratory infections on the index admission, or ICU length of stay (Table 3). However, those who did not undergo primary PT had significantly longer lengths of hospital stay (median 29.5 days [IQR 19–60] vs 18 days [16–32], p = 0.02). There was also a trend toward increased need for airway evaluations on the index admission among patients in the EA group compared to those in the EA + PT group (n = 11, 26% vs n = 2, 10%, p = 0.19). The rates of various post-operative complications, such as vocal fold movement impairment, subglottic stenosis, chyle leak, and esophageal leak were relatively low and similar between the two groups (Table 3).

All infants underwent an esophagram post-operatively prior to initiation of feeds. The esophagus was noted to be to the right of midline on the esophagram in the majority of infants who underwent posterior tracheopexy (86%), while it remained midline in the majority of infants (81%) who did not get a PT upfront (Fig. 2).

3.2. Clinical outcomes within the first year of life

Median follow up for the entire cohort was 28.5 months (IQR 17–45 months), and was not significantly different between groups. Infants in the EA + PT Group were significantly less likely to have respiratory infections requiring hospitalization within the first year of life (0% vs 26%, p = 0.01) and less likely to have episodes of blue spells (0% vs 19%, p = 0.04) (Table 4). Though not statistically significant, those who underwent primary PT were less likely to have had a respiratory infection (at least one) not requiring hospitalization (n = 5, 23.8% vs n = 20, 47.6%, p = 0.06), and were less likely to have parent-reported episodes of noisy breathing, wheezing, or stridor (n = 6, 28.6% vs n = 20, 47.6%, p = 0.11) when compared to those in the EA Group. There was no difference between the groups in the frequency of esophageal stricture during the first year of life, and particularly no infant in the EA + PT Group was noted to have dysphagia attributable to the rightward location of their esophagus (rotational esophagoplasty). Additionally, no infant in either group was noted to have recurrent or acquired TEF on subsequent follow-up. Lastly, infants who underwent primary PT demonstrated significantly improved weight-for-age z scores at

Table 3
Post-operative outcomes and complications.

	EA + PT Group (n = 21)	EA Group (n = 42)	p-value
Post-op neuromuscular blockade (paralysis), n (%)	6 (28.5%)	11 (26.2%)	0.84
Days on paralysis (if applicable), median (IQR)	3 (2–3)	3 (2–5)	0.45
Time to extubation, median (IQR), days	2 (2–5)	3 (2–6)	0.33
Need for reintubation within 72 h post-extubation, n (%)	1 (4.8%)	5 (11.9%)	0.65
Need for PPV within first week post-extubation, n (%)	4 (19.0%)	11 (26.2%)	0.75
Post-op respiratory infection on index admission, n (%)	1 (4.8%)	5 (11.9%)	0.65
Chyle leak	0 (0%)	0 (0%)	1.00
Esophageal leak	2 (9.5%)	2 (4.8%)	0.59
Esophagus location in relation to airway (E-gram):			
Midline, n (%)	3 (14.3%)	34 (81.0%)	<0.001
Rightward, n (%)	18 (85.7%)	8 (19.0%)	<0.001
Vocal fold movement impairment, n (%)	3 (14.3%)	7 (16.7%)	1.00
Subglottic stenosis, n (%)	0 (0.0%)	3 (7.1%)	0.55
Need for post-op airway eval on index admission, n (%)	2 (9.5%)	11 (26.2%)	0.19
Discharged on O2 support, n (%)	0 (0.0%)	2 (4.8%)	0.55
ICU LOS, median (IQR), days	9 (4.5–17.5)	11 (7.8–35)	0.08
Hospital LOS, median (IQR), days	18 (16–32)	29.5 (19–60)	0.02

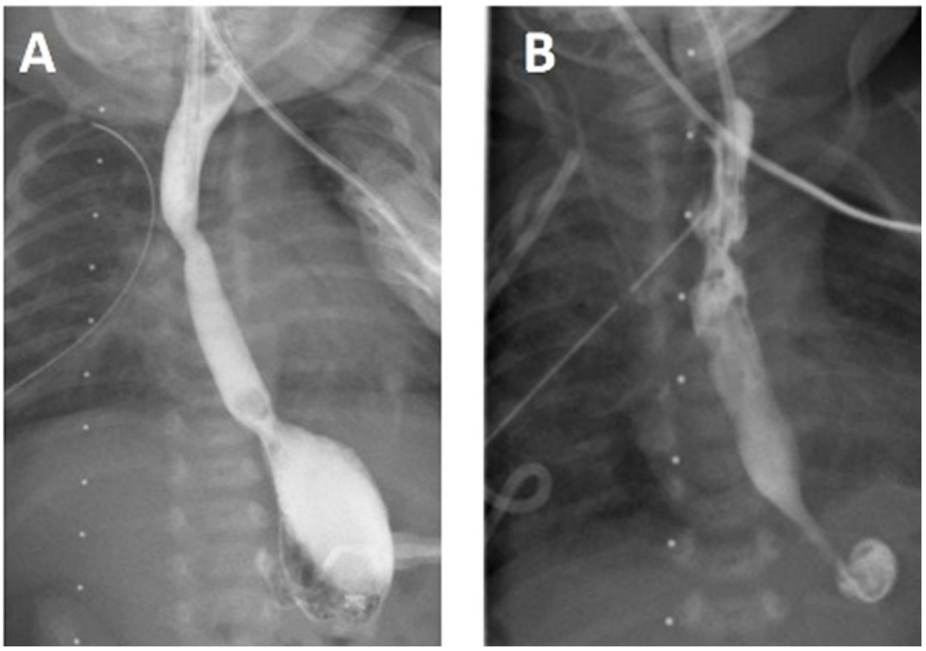


Fig. 2. Representative esophagrams highlighting (A) the rightward location of the esophagus in EA + PT Group vs (B) the more midline location of esophagus in the EA Group.

12 months of age, with a median Z-score of 0.24 (IQR -0.91 to 0.71) vs −1.02 (IQR -1.74 to −0.61) in the group who did not undergo PT ($p < 0.001$).

3.3. Need for delayed posterior tracheopexy

Of the 42 infants in the EA Group, 10 (24%) developed severe TBM symptoms and underwent tracheopexy during the first year of life, whereas no infant in the EA + PT Group needed additional airway surgery ($p = 0.01$). Beyond the first year of life, with a comparable median follow-up between the two groups, four infants in the EA Group progressed to needing either anterior or posterior tracheopexy, while only one infant in the EA + PT Group did (Fig. 3). Most of the airway interventions, when indicated, were thus performed within the first year of life.

4. Discussion

This is the largest study to evaluate the impact of incorporating posterior tracheopexy at the time of newborn EA repair on respiratory and nutritional outcomes. We demonstrate that primary PT, when performed by surgeons experienced with the procedure, can be performed safely in the neonatal period without increasing operative time or complications. We show that incorporating PT at the time of newborn EA repair was associated with a significantly reduced incidence of respiratory infections requiring hospitalization, as well as reduced incidence of blue spells within the first year of life. In fact, no infant in the group who underwent PT required hospitalization for respiratory infection or reported blue spells within the first year of life. We also show significantly improved weight-for-age z scores at 12 months of age for those undergoing PT. Furthermore, among infants who did not undergo PT at the initial operation, we show that approximately 25% of them developed severe tracheobronchomalacia symptoms that resulted in subsequent tracheopexy within the first year of life.

While there is greater experience and data for PT performed secondarily in infants and children with TBM who have previously undergone EA repair, this study focuses on newborns who underwent the procedure at the time of initial EA/TEF repair. Indeed, most infants and children in the former cohort are referred to centers with experience in complex esophageal and airway surgery, whereas neonates in the latter group cannot realistically be

Table 4
Respiratory and feeding outcomes within first year of life.

	EA + PT Group (n = 21)	EA Group (n = 42)	p-value
Length of follow-up, median (IQR), months	30 (14.5–41.5)	27.5 (17.2–48)	0.75
Need for post-EA repair bronchoscopy (after hospital discharge)	11 (52.4%)	23 (54.8%)	0.70
Recurrent/acquired TEF	0 (0.0%)	0 (0.0%)	1.00
Need for delayed tracheopexy within first year of life	0 (0.0%)	10 (23.8%)	0.01
Need for delayed tracheopexy beyond first year of life	1 (4.8%)	4 (9.5%)	0.35
Respiratory tract infections (at least once), n (%)	5 (23.8%)	20 (47.6%)	0.06
Respiratory infections requiring hospitalization, n (%)	0 (0.0%)	11 (26.2%)	0.01
Blue spells, n (%)	0 (0.0%)	8 (19.0%)	0.04
Aspiration events, n (%)	4 (19.0%)	12 (28.6%)	0.54
Oxygen support dependence, n (%)	0 (0.0%)	3 (7.1%)	0.55
Barky cough, n (%)	9 (42.9%)	24 (57.1%)	0.21
Noisy breathing/Stridor/Wheezing, n (%)	6 (28.6%)	20 (47.6%)	0.11
Weight-for-age Z scores at 12 months (IQR)	0.24 (-0.91 to 0.71)	−1.02 (-1.74 to −0.61)	<0.001

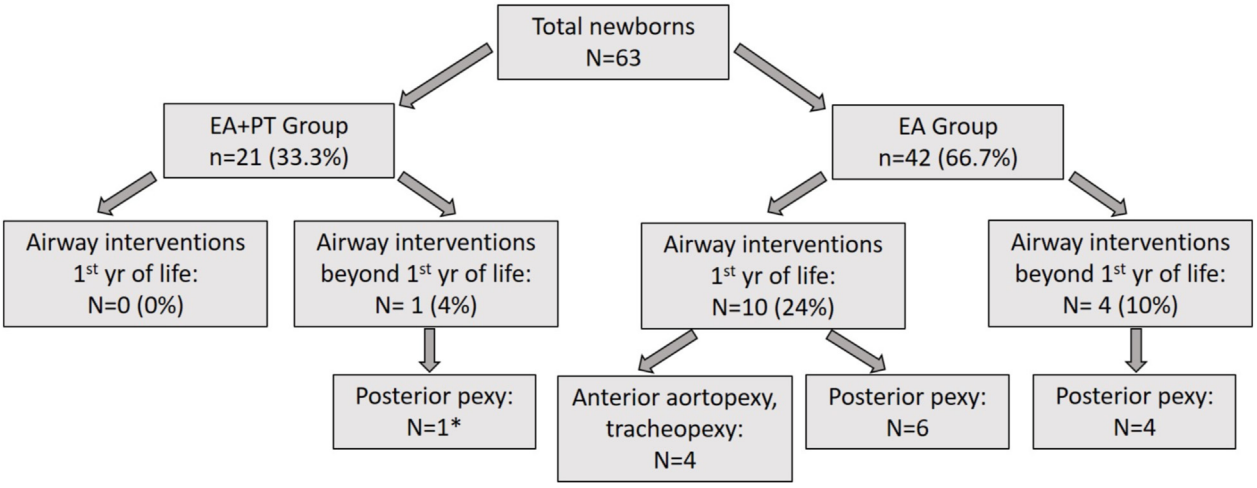


Fig. 3. Distribution of delayed airway interventions within and beyond the first year of life.
* A single patient in the EA + PT group required repeat PT at 19 months of age due to persistent TBM symptoms. The patient's interrupted IVC precluded division of the azygous vein at the index operation, which possibly contributed to suboptimal tracheopexy.

transferred to subspecialty centers. Thus, the idea of widespread implementation of PT for newborns with EA/TEF may be problematic. We do not yet advocate for this as we believe that the technical execution of this procedure and the potential risks must all be balanced with the perceived and demonstrated benefits. As of now, this procedure should probably only be considered in centers with established expertise with PT and surgical management of tracheomalacia. Future work from our team will entail the development of mentoring and proctoring pathways to safely expand this technique to other centers.

We believe posterior tracheopexy should be performed with flexible bronchoscopic visualization in order to assure accurate suture placement. Performing flexible bronchoscopy through endotracheal tubes smaller than size 3.5 is challenging and requires an experienced anesthesia team. If these resources are lacking, safely performing PT for newborns becomes challenging. We do not have a weight limit below which we would defer this procedure, but we recognize that risk for airway injury may increase with smaller patient size. In general, for neonates in this study who were intubated with an ETT size 3 or smaller, suture placement was assessed with intermittent bronchoscopic visualization, if possible. The inability to reliably perform intraoperative bronchoscopy due to patient factors such as size or instability should preclude PT in those neonates.

While no negative consequences of performing PT were observed in this cohort, the various associated risks for the neonate must nevertheless be acknowledged. Circumferential mobilization of the esophagus increases risk of injury to the recurrent laryngeal nerves. However, inadequate mobilization can result in luminal narrowing or angulation, a term we have described as posterior tracheopexy related esophageal stricture (PTRES) [14]. To adequately perform PT, the anterior longitudinal ligament of the spine must be exposed, the process of which requires dissection in the expected location of the thoracic duct, placing it at risk for inadvertent injury. More extensive esophageal dissection could theoretically result in greater devascularization, potentially compromising blood supply to a healing anastomosis and risking esophageal leak or stricture. There were no infants in the EA + PT Group during the study period who had a chyle leak, esophageal leak, esophageal stricture, or PTRES, and the rate of vocal fold movement impairment was similar between the two groups (in the setting of universal pre- and post-operating screening and routine use of intraoperative recurrent laryngeal nerve monitoring) [15–19].

Furthermore, as important as it is to consider performing PT at the time of newborn EA repair, it is just as important to recognize when not to do so. We would not recommend doing a PT in neonates with a midline or anterior descending aorta (unless a descending aortopexy is concurrently performed). Previous work at our institution demonstrated that performing PT without descending aortopexy risks compression of the left mainstem bronchus in this subset of patients [20]. As most newborns with Type C or D EA/TEF do not undergo pre-operative cross-sectional imaging to assess anatomic position of the aorta, the decision to forgo a PT at the index operation would have to be made based on intraoperative findings. No neonate in this cohort underwent descending thoracic aortopexy at the time of EA repair, and none of the infants who did undergo PT as neonates were noted to have left mainstem bronchus compression postoperatively. Another scenario to be mindful of is the case of an aberrant right subclavian artery that courses posteriorly to the trachea. If a PT is truly needed, division with or without reimplantation of the aberrant subclavian artery should be considered.

Limitations to our study include the retrospective and non-randomized study design. We also acknowledge the selection bias inherent in this study as neonates were essentially chosen for primary PT based on bronchoscopy findings and surgeon preference. Neonates in the group without PT had significantly lower birth weight (and non-statistically significant greater proportion of prematurity), which may contribute to the longer length of hospital stay noted in this group. As previously noted, there was a greater prevalence of moderate to severe TBM in the EA + PT Group. It is possible that surgeons considering PT would be more likely to perform a dynamic airway exam and hence more likely to accurately identify and classify the severity of TBM. Furthermore, all PTs were performed by experienced subspecialized surgeons, raising the issue of generalizability. It is likely that our threshold for operative intervention for TBM is lower than that of others, thus impacting the high rate of delayed tracheopexy seen in our follow-up period. Nonetheless, others have also reported rates of ALTE's of up to 22% in the first year of life in EA/TEF patients, along with significant risk of ALTE recurrence with non-tracheopexy management approaches (e.g. esophageal dilation, fundoplication, aortopexy) [21]. None of our infants who required delayed

tracheopexy due to ALTE events had a recurrent ALTE. Indeed, our experience as a referral center for surgical management of TBM has given us the perspective on the benefits that come with breaking the cycle of respiratory morbidity that accompanies TBM in this patient population. Despite these limitations, however, it is worth noting that the study period was narrowed to the most recent five year period where our institutional practice has been for subspecialized surgeons to perform the majority of newborn EA/TEF repairs in a standardized manner. As a result, almost half of the neonates who underwent EA repair without PT were also operated on by the same group of subspecialized surgeons.

Our study demonstrates that the majority of delayed airway interventions (67%) were performed within the first year of life (vs 33% beyond the first year of life). For infants in the EA Group who progressed to need for tracheopexy within the first year of life, surgical planning became complex. Based on the degree of symptoms and time elapsed since initial EA/TEF repair, some of these infants ($n = 4$) underwent sternotomy for anterior aortopexy and anterior tracheopexy first. The rationale for this involved balancing bronchoscopic findings and/or concomitant clinically significant congenital cardiac defects with the risks of a redo thoracotomy too soon after the index repair. Two of these infants continued to struggle after anterior airway interventions, requiring close follow-up, aggressive outpatient medical management, and eventually required posterior tracheopexy via redo thoracotomy, one within the first year of life and one at two years of age.

With respect to improved weight-for-age z scores, it has previously been published that neonates with tracheomalacia have increased work of breathing compared to neonates without TBM [22]. It is thus not surprising that infants with TBM, who are putting in greater effort to breathe, may have greater rates of feeding difficulties and compromised ability to maintain their ideal body weight. This increased work of breathing translates into increased energy expenditures, something that needs to be taken into account as part of their nutritional assessment and optimization strategy [23]. The anatomic differences for esophageal position with and without PT likely also affect work of breathing specifically during feeding when esophageal distension can impact posterior tracheal compression and respiratory efficiency while swallowing.

With such compelling reduction in respiratory morbidity among neonates in the EA + PT Group, prospectively studying the role for PT in a randomized setting may be warranted. Continued investigation is needed to demonstrate the durability of posterior tracheopexy on subsequent bronchoscopic evaluations. While the vast majority of our newborn repairs were done via the open technique, extending this study to the minimally invasive approach also warrants future investigation. However, our group and other centers have reported on thoracoscopic PT, including in neonates, with encouraging results [11–13]. Currently reported techniques of thoracoscopic PT often utilize simple sutures rather than pledgeted mattress sutures; this technical difference should also be studied in terms of effectiveness and durability. Further follow-up and subset analysis is also needed to demonstrate whether performing a delayed tracheopexy resulted in reduced respiratory morbidity or improved nutritional outcomes for that particular cohort.

5. Conclusion

Incorporating a posterior tracheopexy during the newborn EA/TEF repair is associated with significantly reduced respiratory morbidity and significantly improved weight-for-age z scores within the first year of life. While continued evaluation is necessary, we recommend that a prophylactic posterior tracheopexy, under bronchoscopic guidance, be considered at the time of initial EA/TEF repair in centers with experience with PT and surgical management of TBM.

Conflict of interest

The authors have no sources of financial support or conflicts of interest that are relevant to this work.

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