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REVIEW ARTICLE

Anesthetic management of congenital tracheoesophageal fistula

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Summary

This article reviews (a) risk factors and preoperative considerations of the patient with tracheoesophageal fistula, (b) anesthetic management, including (i) airway management, (ii) induction of anesthesia and monitoring and (iii) postoperative disposition, (c) considerations for concomitant congenital heart disease, (d) considerations for thoracoscopic repair and (e) long-term outcomes and considerations of the patient with repaired esophageal atresia/tracheoesophageal fistula.

Introduction

Advances in surgical practice and neonatal intensive care mean that neonates with congenital esophageal atresia/tracheoesophageal fistula (EA/TEF) often present with minimal lung pathology and excellent lung compliance. Many pediatric anesthesiologists routinely paralyze these patients following demonstrated adequacy of bag-mask ventilation and position the endotracheal tube (ETT) well above the fistula, without compromising ventilation. Perioperative mortality continues to fall, and yet, these repairs can be fraught with complications. Critically, anesthesiologists must recognize those patients at additional risk: those with coexisting complex congenital heart disease (CHD), weight < 2 kg, poor pulmonary compliance, large, pericarinal fistulas and those scheduled for thoracoscopic repairs. Rigid bronchoscopy to characterize airway anatomy helps guide advanced airway strategies and decisions regarding postoperative disposition.

Risk factors and preoperative considerations

Congenital EA/TEF has an incidence of 1 : 2500–3000 live births and is described by two major anatomic classification systems: Gross and Vogt (Figure 1) (1). The common form is EA combined with a distal fistula (C/IIIb). The surgical management is right thoracotomy or thoracoscopic primary repair; occasionally, staged repair is indicated for long-gap atresia. Isolated TEF (E/H-type) is managed with a right cervical dissection (occasionally thoracotomy or thoracoscopic surgery if the fistula is low) (2,3) or with minimally invasive, rigid bronchoscopic application of tissue adhesive or fibrin glue (also indicated for recurrent fistulas). Low success rates with endoscopic therapy mean that definitive closure often requires a second attempt (4,5). Isolated EA (A/II) is managed by gastrostomy, with repair in 2–3 months. Without a fistulous connection to the trachea, these patients pose fewer challenges.

Unlike many anomalies, TEF does not lend itself to prenatal diagnosis (6). Nonspecific ultrasound signs (polyhydramnios, absent or small stomach bubble) have a positive predictive value of 44%, leaving clinical suspicion the key to diagnosis. Clinicians watch for excessive salivation, choking with feeds and the inability to pass a suction catheter more than 9–10 cm into the esophagus. A radiogram of thorax and abdomen is requisite because, typically in EA, the presence of gas below the diaphragm indicates TEF. Although isolated TEF is often diagnosed in the newborn period, a high index of suspicion is required to recognize cyanosis with feeds, recurrent lower respiratory tract infections

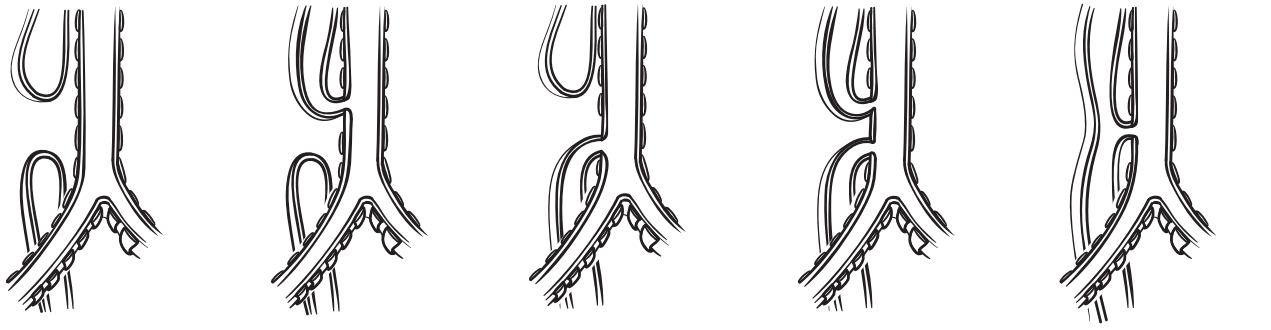

Figure 1 Anatomic classification and incidence.

and intermittent abdominal distension and to obtain radiologic or endoscopic confirmation (7,8).

Early experience with repair of EA/TEF was characterized by high mortality rates. Waterston introduced prognostic classification in the 1960s (Table 1) (9–11). His three determinants were weight, pneumonia and associated congenital anomalies. In the 1990s, Spitz recognized the congenital anomalies affecting prognosis were cyanotic heart disease requiring palliative or corrective surgery, or noncyanotic heart disease requiring medical or surgical management. Pneumonia was so rare as to be removed from the classification. Okamoto used stepwise logistic regression and receiver-operating characteristic (ROC) curves of birth weight to statistically extrapolate the 'at-risk' weight group as <2 kg. A less-than-2-kg baby with congenital heart disease still has a survival rate of 27%, whereas greater-than 2-kg neonates without cardiac disease should have survival rates approaching 100%.

Coexisting congenital abnormalities occur in 50% of patients with EA/TEF (Table 2) (1,12), more common in isolated EA (65%) than in isolated TEF (10%). The prevalence of congenital heart disease necessitates a preoperative electrocardiogram and echocardiogram in all patients. Congestive heart failure must be medically optimized. Diaz *et al*., using retrospective case–control

Table 1 Prognostic classification of EA/TEF by Waterston, Spitz, Okamoto

Prognostic Scheme	Class	Birth Weight	Anomaly	Survival Rate (%)
Waterston 1962	A	≥2.5 kg	Nil	95
	B	≥2.5 kg	Lobar pneumonia mild – moderate congenital anomaly (i.e., limb anomalies, cleft palate, atrial septal defect, patent ductus arteriosus)	68
		1.8–2.5 kg	Nil	
	C	1.8–2.5 kg	Severe pneumonia Severe congenital anomaly (i.e. transposition of the great arteries, other bowel atresia, renal insufficiency), or combinations of moderate anomalies	6
		<1.8 kg	Nil	
Spitz 1994	I	≥1.5 kg	Nil	97
	II	≥1.5 kg	Major cardiac anomaly (i.e. cyanotic CHD or noncyanotic CHD requiring medical or surgical treatment for heart failure)	59
		<1.5 kg	Nil	
	III	<1.5 kg	Major cardiac anomaly	22
Okamoto 2009	I	≥2.0 kg	Nil	100
	II	<2.0 kg	Nil	81
	III	≥2.0 kg	Major cardiac anomaly	72
	IV	<2.0 kg	Major cardiac anomaly	27

Table 2 Congenital abnormalities associated with EA/TEF

Organ System	Incidence (%)	Examples
Cardiac	29	VSD, PDA, tetralogy of Fallot, ASD, right-sided aortic arch
Gastrointestinal	14	Duodenal atresia, imperforate anus, malrotation, pyloric stenosis, omphalocoele
Genitourinary	14	Renal agenesis, hypospadius, horseshoe/polycystic kidney, ureteric/urethral abnormalities
Musculoskeletal	10	Radial limb abnormalities, polydactyly, lower limb defects, hemivertebrae, rib defects, scoliosis
VATER syndrome (VACTERL)	10	Vertebral, anorectal, tracheoesophageal, renal or radial anomalies (expanded to include cardiac and limb defects) other associated syndromes include: CHARGE (coloboma, heart defects, atresia choanal, retarded growth and development, genital hypoplasia and ear deformities), Potter's (pulmonary hypoplasia, bilateral renal agenesis, characteristic facies of intrauterine compression), SCHISIS (omphalocoele, cleft lip and/or palate, genital hypoplasia)
Respiratory	6	Tracheo-bronchomalacia, pulmonary hypoplasia, tracheal agenesis/stenosis, tracheal upper pouch
Genetic	4	Trisomy 21, Trisomy 18, 13q deletion

VSD, ventricular septal defect; PDA, patent ductus arteriosus; ASD, atrial septal defect.

data comparing 26 patients with complex CHD to 27 patients without significant cardiac abnormalities, demonstrated significantly more perioperative critical events in patients with complex CHD (but no different perioperative adverse events between ductal-dependent cardiac lesions compared to other cardiac conditions) (13). Considering the risk of vertebral anomalies, all neonates with a sacral dimple would benefit from lumbar ultrasound, particularly if planning a caudal catheter.

Pulmonary insufficiency has important implications for anesthetic management. Poor lung compliance increases gastric distension and ventilatory compromise. Major etiologies include respiratory distress syndrome (RDS) complicating prematurity and aspiration pneumonia. Prematurity with EA/TEF has an incidence of 30% (12,14); however, improvements in neonatal intensive care, including the use of maternal steroids and exogenous surfactant, have decreased RDS. Delayed diagnosis of a fistula contributes to the development of pneumonia. Aspiration is minimized by suction of the blind esophageal upper pouch by a Replogle tube (double-lumen, radio-opaque tube for continuous suction and irrigation) and early surgery. In the patient with isolated TEF, institution of nasojejunal feeds may permit preoperative resolution of aspiration pneumonia (7). In the neonate with a distal fistula and pulmonary pathology resulting in decreased pulmonary compliance, expect challenging ventilation. To facilitate mechanical ventilation, the priority must be ligating the fistula, performed via thoracotomy or transabdominally (1,14). Primary repair of the EA and division of the fistula may proceed if the child stabilizes or may be deferred until resolution of lung pathology (or within 8–10 days for fear of re-fistulization following a temporizing ligation).

Anesthetic management

Airway management

Neonates with TEF are at risk of gastric distension and potentially pneumoperitoneum (treated by emergent needle decompression of the left upper quadrant of the abdomen). Traditionally, EA/TEF was managed by a staged procedure, and anesthetic texts continue to mention preoperative gastrostomy. While gastrostomy obviates life-threatening gastric rupture, the placement of a gastrostomy may cause further ventilatory instability, as it provides a low-pressure leak via a 'bronchocutaneous' fistula, with resultant ineffective ventilation (15,16). During spontaneous ventilation, there is little gastric insufflation, even with poor lung compliance; institution of positive pressure ventilation imposes this risk, particularly in the setting of pulmonary pathology. When faced with inadequate ventilation intraoperatively, the anesthesiologist must entertain a wide differential diagnosis that includes gastric distension, but also other common causes including ETT obstruction (13,17).

The potential for two fistulas, and the complications from an unrecognized, unrepaired fistula, means that many surgeons begin with rigid bronchoscopy to define the anatomy, prior to performing the repair (18,19). Although the provision of anesthesia for rigid bronchoscopy in a spontaneously ventilating neonate

is challenging, this is an opportunity to characterize the airway. Firstly, these patients are at risk of concomitant airway anomalies, including tracheo-bronchomalacia, tracheal upper pouch and tracheal stenosis/agenesis, which may alter the decision to proceed with muscle relaxation (1,20,21). Secondly, the position of the fistula affects management options: it may be difficult to intubate beyond a pericarinal fistula. Holzki *et al*. published the bronchoscopic findings of 113 neonates: in 67%, the TEF was >1 cm above the carina, 22% <1 cm above the carina and 11% below the carina (22). Thirdly, in a retrospective review of 61 cases at Children's Hospital, Oakland, no patients with a small fistula (as opposed to large >3 mm fistula) presented a ventilation challenge (14). If bronchoscopy reveals a large fistula, particularly in the setting of decreased lung compliance, the anesthesiologist must have particular concern regarding gastric distension. Numerous airway strategies have been attempted to ensure adequate mechanical ventilation: one-lung ventilation, tracheal intubation distal to the fistula, Fogarty catheter occlusion and fiberoptic tracheoscopy.

Case reports document the successful use of one-lung ventilation in term and premature neonates (34 weeks, 1.5 kg) (23,24). An inhalational agent in 100% O_2, supplemented by fentanyl 1 mcg·kg^{-1}, induction agent and muscle relaxant, facilitates ETT position in the left mainstem bronchus for the duration of fistula ligation, whereupon the tube may be withdrawn to the trachea for the esophageal anastomosis. One-lung ventilation has the advantage of a still operative field for thoracotomy or thoracoscopic repairs. The anesthesiologist should confirm aortic arch position as $\sim$2.5% will have a right-sided aortic arch and require a left thoracotomy (25). Krosnar *et al*. (12) described a case managed with one-lung ventilation that was complicated by left lung atelectasis and required postoperative reintubation. Whether the atelectasis was related to bronchial edema secondary to traumatic intubation, or coincident bronchomalacia/stenosis was unknown.

Intubation distal to the fistula was first described by Salem in 1973 (26). An ETT without side hole is positioned above the carina with the bevel anterior to maximize occlusion of distal fistula and ventilation of both lungs. Several groups in the literature have reported success with this method (12,14), but it is not without danger. Unintended ETT migration, either into the underventilated left mainstem bronchus or catastrophically into the fistula as described in a recent case report (27), can occur. Use of a flexible fiberoptic bronchoscope to guide ETT fixation can mitigate these risks and is recommended.

Occlusion of the fistula with a Fogarty arterial embolectomy catheter has been used effectively in patients with large, distal fistula and poor lung compliance (14,28,29). During rigid bronchoscopy, the catheter may be directed by the surgeon to occlude the fistula. Other options include blind or fiberoptic placement either alongside or within an ETT. Nevertheless, Fogarty catheters are challenging to place and may potentially dislodge, causing central airways obstruction (30). They may have a role as a 'Plan B': a 2 Fr (4-mm balloon) or 3 Fr (5-mm balloon) kept on hand during rigid bronchoscopy, to be placed in large, pericarinal fistulas when the tracheas are not amenable to intubation beyond the fistula.

A group from Zurich described extensive experience in 47 neonates weighing 1.1–3.8 kg (31–42 weeks of gestation) with tracheoscopy as an alternative to rigid bronchoscopy (17). Using 2.0, 2.4 and 2.8 mm OD flexible fiberoptic bronchoscopes, three endoscopic tracheal examinations were undertaken via the ETT per case. Firstly, airway anatomy was delineated (occasionally with 5–10 cm H_2O PEEP applied to 'open the orifice'). Later, transillumination of the fistula permitted rapid surgical control (31). Finally tracheoscopy assessed postoperative bleeding, secretions, and tracheomalacia. Tracheoscopy has the advantages of a secured airway and muscle relaxation, which minimizes anesthetic cardiovascular depression. None of the patients had respiratory deterioration despite mechanical ventilation via an ETT positioned well above the fistula (albeit, the surgical time to ligation was minimized by transillumination). There were two cases of inadvertent extubation. Despite the success of this technique in the hands of a skilled bronchoscopist, there are two critiques: tracheoscopy through an ETT might miss a high fistula and the use of a fiberoptic bronchoscope in combination with a small ETT and 'orifice-opening' pressures may cause a pneumothorax (32).

Induction of anesthesia and monitoring

Traditionally, the maintenance of spontaneous ventilation has been advocated in EA/TEF. Considering the possibility of additional airway anomalies and large (>3 mm), pericarinal fistulas, this remains a safe choice until more information about the airway is available. While maintenance of spontaneous ventilation during rigid bronchoscopy is challenging, the addition of propofol infusion to an inhalational technique is helpful. Nonetheless, most cases will do well with intravenous induction, including institution of muscle paralysis following demonstrated ease of bag-

mask ventilation without gastric distension, as established by Deanovic (17). Those patients with poor pulmonary compliance, being at greater risk of gastric insufflation, should be approached conservatively.

In addition to standard noninvasive monitors, invasive arterial monitoring is indicated in patients with comorbid disease, particularly complex congenital heart disease or pulmonary disease, and in patients undergoing thoracoscopic surgery.

Postoperative disposition

Postoperative tracheal extubation is determined by complications related to airway anatomy: tracheobronchomalacia, recurrent laryngeal nerve injury, overlooked fistula and recurrent fistula (33,34). Although tracheo-bronchomalacia is present in 75% of pathologic specimens, clinically relevant malacia occurs in 10–20%, rarely requiring aortopexy (35,36). Recurrent laryngeal nerve injury also occurs. Diaz *et al*. (13) report 15 extubations of 38 neonates not intubated preoperatively, two of which required reintubation for vocal cord paresis. Overlooked upper pouch fistula and fistula recurrence (3–15% incidence with surgery, ~50% with endoscopic repair) should be suspected in patients difficult to wean from mechanical ventilation (4). Enteral nutrition is usually commenced via a transanastomotic feeding tube, which must not be displaced if tracheal extubation is planned (37).

Opioid sparing techniques, either thoracoscopic surgery or regional anesthesia, facilitate earlier tracheal extubation by reducing the risk of apneas. Thoracoscopic procedures have earlier extubation times (37.6 vs 54 h) and discharge from intensive care units (2.75 vs 3.4 days) than thoracotomies, with similar postoperative complication rates (12,38,39). Caudal catheters, threaded to the thoracic region, are utilized in some centers to control postoperative pain (40). A variety of approaches have been advocated for positioning the caudal catheter in the thoracic region, from ultrasound imaging to the Tsui technique (41). Confirmation of catheter position with fluoroscopy remains definitive (42). Recommended maximum dose for bupivacaine infusion in neonates is 0.2 $mg \cdot kg \cdot h^{-1}$. Immature cytochrome P450 pathways in neonates mean rising plasma concentrations of bupivacaine after 48 h, after which time prolonged infusions must be reduced, perhaps by as much a 40% (43). Avoidance of regional anesthesia with its corresponding decrease in systemic vascular resistance is warranted in patients with coexisting congenital heart disease such as hypoplastic left heart syndrome (HLHS).

Anesthesia for concomitant complex congenital heart disease

Management of neonates with TEF and coexisting complex CHD benefits from a team approach. Whereas TEF ligation may be an emergent procedure in neonates failing mechanical ventilation, EA/TEF repair is an urgent operation to minimize aspiration pneumonia and begin enteral nutrition. Nonetheless, a few non-ductal-dependent cardiac lesions cause significant physiologic impairment in the first days of life; thus, there is no reason to delay repair (44). In ductal-dependent patients, the options include proceeding with EA/TEF repair while maintaining the prostaglandin infusion (PGE_1) or proceeding with a palliating procedure. Walker *et al*. (40) describe their management of four EA/TEF repairs in patients with congenital heart disease (HLHS, tetralogy of Fallot, truncus arteriosus misdiagnosed as a VSD on echocardiogram). Anesthesia in these patients was induced using spontaneous inhalation of 5% sevoflurane in either air or 100% O_2 to facilitate rigid bronchoscopy. All had invasive arterial and central venous pressure monitoring. In Diaz's publication, 60% of patients with complex CHD had invasive monitoring during TEF repair, typically umbilical artery and vein catheters (13). If the team decision is to proceed with cardiac palliation prior to fistula ligation in an unintubated patient, the anesthesiologist may prefer to involve an otolaryngologist. Rigid bronchoscopy to delineate airway anatomy may prevent intubation of a tracheal upper pouch or other potential airway disaster in the catheterization laboratory.

Diaz *et al*. (13) describe six patients with duct-dependent circulation for EA/TEF repair maintained intraoperatively on PGE_1; all were intubated preoperatively, had central venous pressure monitoring and access and most had invasive blood pressure monitoring. Despite high long-term mortality rates in the ductal-dependent groups, there was no significant difference in intraoperative critical events between the ductal-dependent and non-ductal-dependent groups. Indeed, Diaz suggests early TEF repair in patients with pulmonary overcirculation, prior to the fall in pulmonary vascular resistance.

A case report describes the management of term 2.26 kg neonate with prenatally diagnosed HLHS and clinically diagnosed EA/TEF (45). To facilitate ventilation during a planned Norwood procedure, on day four, the patient was taken to theater for expeditious rigid bronchoscopy, thoracotomy with fistula ligation and gastrostomy. Although the patient was briefly exposed to 100% O_2 for intubation, the FiO_2 other-

wise was titrated upwards of 0.21 only to maintain SpO_2 within 70–80% range. Despite manual ventilation during compression of the right lung, $PaCO_2$ rose to 80 mmHg. Hemodynamic instability occurred with improved ventilation at the conclusion of the case. An epinephrine infusion was required in the short term and then weaned. This case illustrates how physiologic changes experienced during the TEF repair compliment the management of pulmonary and systemic circulations in a ductal-dependent patient.

Anesthesia for thoracoscopic repair of EA/TEF

The anesthetic management of thoracoscopic EA/TEF repair lends another level of complexity to the case (46–49). Since 2001, over 50% of EA/TEF cases have been managed thoracoscopically at the Royal Hospital for Sick Children, Edinburgh (12). Krosnar *et al.* presented a review of eight cases, all stable preoperatively with minimal O_2 requirement, nil or mild cardiac anomalies (PDA, PFO, small perimembranous VSD) and nil or mild prematurity (34 weeks 5 days, 36 weeks 5 days). Surgical duration varied between surgeons, in the range of 130–220 min. All neonates were monitored with an arterial line and had large bore venous access and an inhalational induction with or without muscle relaxation. The ETT placement was confirmed bronchoscopically, distal to the fistula in seven and in the left mainstem bronchus in the eighth (as previously discussed, this patient's postoperative course was complicated by left upper lobe atelectasis). The neonates were maintained on inhalational agent and opioid with intermittent positive pressure ventilation or manual ventilation. Surgical insufflation of CO_2 to 5 mmHg collapsed the right lung, and patients desaturated to 84–91% and mostly required 100% O_2. Blood loss was minimal. The seven were taken tracheally intubated to PICU on morphine infusions of 20–40 $mcg{\cdot}kg{\cdot}h^{-1}$ following local anesthetic infiltration of the port sites; the eighth was extubated and required emergent reintubation following a respiratory arrest 12 h later. During one-lung ventilation, endtidal CO_2 is falsely low, and arterial blood sampling permits monitoring of ventilation; nonetheless, a sudden increase in $ETCO_2$ should alert the anesthesiologist to possible surgical trauma and entrainment of thoracoscopic gas.

Long-term outcomes and considerations of patients with repaired EA/TEF

The anesthetic concerns in a patient with repaired EA/TEF include dysphagia, gastro-esophageal reflux disease (GERD), tracheomalacia, bronchial hyperreactivity and occasionally scoliosis (33). Patients return to operating theater for dilation of esophageal strictures. Dysphagia and gastrointestinal problems lead to hospitalization in 65% of patients prior to ten years of age. GERD occurs in 35–58% of children and persists into adulthood. Respiratory complications, including recurring bronchitis (most common in infancy) and wheezing, are multi-factorial: secondary to tracheomalacia, GERD and recurrent fistulas. An association with asthma is unclear, because bronchial hyper-reactivity may be attributable to GERD. Chest wall deformities have been attributed to post-thoracotomy syndrome; however, 10% have coexisting vertebral anomalies, which predispose to scoliosis.

Conclusions

Preoperative assessment of the newborn with EA/TEF focuses on delineating complex CHD, impediments to pulmonary compliance, other comorbid disease, weight (< 2 kg) and the demands of the surgical approach. Considering the possibility of concomitant airway anomalies and of large (> 3 mm), pericarinal fistula, it may be prudent to visualize the airway (via rigid or flexible bronchoscopy) prior to muscle relaxation, although in the vast majority of cases, this practice would be uneventful. Whereas positioning the ETT tube in the left mainstem bronchus or distal to the fistula should minimize gastric insufflation and improve both ventilation and the surgical field, the anesthesiologist must be vigilant regarding the catastrophic risk of tube malposition. Fiberoptic bronchoscopy may assist positioning. Fixing the ETT above the fistula has the advantage of avoiding malposition and has been reported to cause no disturbance to mechanical ventilation in 47 patients. Protracted time to fistula ligation, potentially exacerbated by a difficult surgical field, may be a concern in this situation. Patients with ductal-dependent circulation have equivalent intraoperative risk as non-ductal-dependent lesion, provided appropriate anesthetic goals achieved, but have higher morbidity and mortality in the long term related to their cardiac disease. Invasive monitoring is indicated for patients with complex CHD and during thoracoscopic surgery. Postoperative management requires an intensive care setting. Tracheal extubation may be complicated by poor pain control, tracheomalacia and vocal cord paresis among others. Following repair, this population may have significant dysphagia, GERD and bronchial hyperactivity as children and adults.

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