ORIGINAL ARTICLE

Tracheoesophageal fistula in the developing world: are we ready for thoracoscopic repair?

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Abstract

Purpose Tracheoesophageal fistula (TEF) is a bellwether for a country's ability to care for sick newborns. We aim to review the existing literature from low- and middle-income countries in regard to management of those newborns and the possible approaches to improve their outcomes.

Methods A review of the existing English literature was conducted with the aim of assessing challenges faced by providers in LMIC in terms of diagnostic, preoperative, operative and post-operative care for TEF patients. We also review the limited literature for performing thoracoscopic repair in the developing world context and suggest methods for introduction of advanced thoracoscopic procedures including techniques for providing anesthesia to these challenging babies.

Results While outcomes related to technique from LMIC are comparable to the developed world, rates of secondary complications like sepsis and pneumonia are higher. In many areas, repairs are conducted in a staged fashion with minimal utilization of thoracoscopic approach. The paucity of resources creates strain on intraoperative and post-operative management.

Conclusion Clearly, not all developing world contexts are ready to attempt thoracoscopic repair but we outline suggestions for assessing the existing capabilities and a stepwise gradual implementation of advanced thoracoscopy when appropriate.

Keywords Global health · Minimally invasive surgery · Thoracoscopy · Neonatal surgery · Neonatal anesthesia

Introduction

The clinical results of esophageal atresia (EA) have been considered to reflect the level of medicine of an individual country [1]. The fact that EA mortality in low- and middle-income countries (LMIC) is still being reported to range from 30 to 80% is an area for improvement [2, 3]. Some of the obstacles highlighted by LMIC reported literature include delayed diagnosis and/or referral of patients with EA, the lack of continuous suctioning facilities, the higher rates of complications including sepsis and aspiration pneumonia, and a paucity of trained personnel in neonatal anesthesia and critical care.

Challenges in the developing world for TEF repair

Delayed diagnosis of EA has been shown to be an independent negative prognostic factor in LMIC countries [4]. Infants presenting late are at a higher risk of developing aspiration pneumonia [5, 6]. A baby referred from an outside hospital, a history of contrast study, and/or a history of attempts at oral feeding have been associated with aspiration pneumonia and should prompt heightened suspicion and further investigation. Managing aspiration pneumonia with antibiotics prior to surgical intervention has shown to be efficacious in increasing survival [5]. Efforts for these vulnerable babies could include increasing awareness

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of EA among pediatricians and advising against using contrast studies along with establishing prompt referral guidelines. The lack of continuous suction in facilities also contributes to higher rates of aspiration pneumonia [5]. One study suggested compensation by placing the babies in lateral or prone position with repeated suction by oral mucus aspirator or by syringe [7] if infrastructure upgrades are impossible. One can speculate that the frequency of suctioning will depend on the quantity of the oral secretions and that in such a scenario, intensive and one-to-one nursing care is of utmost importance. Another area of concern is perioperative nutritional optimization. While TPN might be the standard in the developed countries, securing an IV access and the availability of TPN is a major hurdle in many developing countries. As in developed countries, patients with long gap esophageal atresia can benefit from placement of a gastrostomy tube before surgical interventions.

Interestingly, leak rates, one of the major complications of EA, do not appear to be higher in LMIC with reports frequently below 20% [2, 5, 8]. And yet, fear of leak instigates a great deal of anxiety for LMIC surgeons to the point of opting to perform staged repair with esophagostomy to avoid this complication [9]. The potential sources for this anxiety are:

(i) Many patients referred for EA repair are malnourished (with delayed presentation) which potentially hinders the healing process [8],

(ii) Lack of neonatal post-operative ventilatory support worries surgeons caring for babies who are at risk for sepsis and the resultant complications.

Staged repair has not been shown to decrease mortality in LMIC as opposed to primary repair [9] and it has even been linked to higher mortality in developed countries [11]. Many methods have been tried as a way to reduce leak rates and have had some success though further studies would be needed before adoption could be recommended, these include:

(i) Placing patients on parenteral albumin infusion postoperatively. One report suggested a decrease leak rates in malnourished patients [8].

(ii) Keeping the patients paralyzed post op for 3–7 days is reported from Japan and Europe [1, 10, 17]. It is hypothesized that this reduces tension on the anastomosis, a risk factor for anastomotic leak.

(iii) Decompressing gastrostomy and feeding jejunostomy can be an alternative to managing major anastomotic leaks [14] and, along those lines, might be utilized as a prophylactic intervention when anticipating a higher risk of anastomotic leak. Obviously, one must balance the high rate of complications associated with feeding jejunostomies in babies when considering this option.

(iv) Administration of glycopyrrolate in patients of anastomotic leak after primary repair of esophageal atresia. The potential benefit is reducing oral secretions, which would help in healing of the anastomotic dehiscence [23].

The most consistently reported major contributor to morbidity and mortality in developing countries is sepsis [2, 3, 5, 8, 9]. Rates of mortality after neonatal surgeries in the developing world from sepsis have been reported as high as 67% making it the most common cause of mortality [15].

A source of sepsis is not always reported in the reviewed literature. It is likely that bacteremia and sepsis in this patient population is multifactorial and related most to postoperative leaks and the sequelae associated with leaks—much more rarely surgical site infections. Given that the leak rate in published series is the same between open and thoracoscopic repair, it is of most importance that each site strengthens its ability to accurately diagnose sepsis and leaks. Some of the limitations for accurate reporting on leak rates are due to lack of adequately trained staff in the neonatal intensive care unit but some areas of opportunity exist in the diagnostic radiology sphere where some hospitals do not have routine access to fluoroscopy.

In spite of sepsis being reported differently, every attempt should be made to decrease the incidence of preoperative pneumonia. *In press* reports of country-wide education on pediatric surgical conditions are just one way to hasten recognition and appropriate transfer of conditions like esophageal atresia.

Resolute efforts in the areas of training the staff on infection control, establishing protocol for routine cleaning of cots and incubators and minimizing thoroughfare in newborn wards are some of the steps that can be taken to address this issue [16].

Lastly, birth weight is still an issue in LMIC. Some authors suggested that much of the literature reported from developed countries advocates abandoning the birth weight parameter from the Waterston classification, as it does not predict survival of patients with EA. Unfortunately, this does not hold true in LMICs [2, 8, 13]. We believe that a case-by-case approach with insight into one's personal and institutional limitations is needed to address birth weight-related mortality and to achieve a balance between speedy intervention and avoidance of causing harm in the interest of expediency. For example, an option to allow for growth and decrease potential morbidity and mortality is performing a fistula ligation with insertion of a gastrostomy tube followed by definitive repair at an appropriate weight if proper options exist for management of the upper pouch secretions. There

are multiple situations at which the above scenario will be appropriate. An unstable neonate unable to tolerate mechanical ventilation due to a large TEF would be better served with an emergent fistula ligation and delayed esophageal repair. It is an appropriate intraoperative decision in patients with long gap esophageal atresia with inability to perform an anastomosis with undue tension. Other indications not infrequently encountered in LMIC are to temporize a neonate born at a facility lacking infrastructure for post-operative management of those patients. Finally, fistula ligation with gastrostomy tube placement can be used with very low birth weight neonates until a target weight is reached. Surgeon experience, non-surgical service support (anesthesia, neonatology, etc.), and equipment availability should determine the optimal weight at a particular institution.

Thoracoscopic repair in the developing world

Although thoracoscopic repair of EA–TEF has been described more than 10 years ago, this approach is performed at only selected centers in the developed countries with an international survey (of developed and developing countries) showing that approximately 94% of respondents prefer an open approach [17]. Reports from Asia and the Middle East [18–22] show that thoracoscopic repair has been attempted but are still on the learning curve and only attempting thoracoscopic repair to infants with birth weights above 1.5 kg. While some studies reported longer operative time (175–230 min) [20, 22], others were within the ranges reported from developed countries (120–150 min) [21]. Technical difficulties and a steep learning curve are frequently cited limiting factors in adopting thoracoscopic repair. Some suggested solutions from surgeons in LMIC are (1) introducing a thoracoscope through a minithoracotomy to learn the videoscopic anatomy and attempting the first few steps of the procedure with thoracoscopic instruments as a first step in the learning curve [21], (2) interdisciplinary team discussion between the pediatric surgery team and the anesthesia team to avoid cases of conversion, (3) mastering advanced laparoscopic suturing skills in other laparoscopic neonatal procedures before performing this procedure [18]. However, it is our opinion that, in the hands of surgeons facile with advanced minimally invasive surgery, with proper safeguards, preparation, and counseling of the patient, it is reasonable to attempt thoracoscopic repair in the right developing world context. Ideally, a surgeon experienced with the approach should be present and each case should be approached with a low threshold for conversion to open.

Anesthetic management

Anaesthetic management during the repair of EA has been shown to significantly affect mortality in EA repair [12]. Up to two-thirds of EA/TEF patients will have one or more chromosomal, isolated cardiac, or VACTERL spectrum anomalies [24], therefore, anesthetic management begins with a thorough history and physical examination, as well as plain radiographs, echocardiogram, renal and spinal ultrasound where available in LMIC. Incidence of critical intraoperative events during repair of EA/TEF is significantly higher in infants with associated cardiac pathology; when compared to nonductal-dependent lesions, presence of ductal-dependent cardiac lesions increase risk of mortality from 10 to 57% [25]. Therefore, identification and preparation for cardiac anomalies are paramount in the child presenting for TEF/EA repair. If advanced diagnostic modalities are not available in LMIC, then attempts should be made to identify cardiac pathology using physical exam and available surrogate examinations.

Airway management often presents the greatest intraoperative challenge during EA/TEF repair. Many EA/TEF patients have decreased lung compliance, often from prematurity or recurrent aspiration pneumonia. Initiation of non-invasive positive pressure ventilation in this patient population can lead to gastric distention. Gastric distention can cause reflux and aspiration of gastric contents, causing pneumonitis. Additionally, the resulting increased intraabdominal pressure decreases venous return, restricts diaphragmatic excursion, and decreases pulmonary compliance. While gastric decompression (either by needle gastrostomy by the surgeon at time of anesthetic induction or by opening a previously placed feeding tube) can prevent life-threatening gastric rupture, it may cause further ventilatory instability by providing a low-pressure leak via a "bronchocutaneous" fisula, with resultant ineffective ventilation [26]. For these reasons, maintaining spontaneous ventilation is preferred during the induction of general anesthesia in this patient population. This can be achieved by volatile anesthetics, or intravenous agents if access has already been secured. If intravenous agents are chosen for induction, great care must be taken to maintain spontaneous ventilation until easy bag-mask ventilation is demonstrated. Many surgeons will perform rigid bronchoscopy to further characterize the airway, to examine the fistula (and check for additional airway abnormalities). This is usually performed after induction but prior to placement of an endotracheal tube. It is safest to maintain spontaneous ventilation for this portion of the procedure. Neuromuscular blockade is usually avoided. If muscle paralysis is desired, it is extremely important to ensure that controlled bag-mask ventilation can be achieved without gastric distention first.

Multiple options exist for intraoperative airway management. Standard intubation and ventilation distal to the fistula can be achieved in many patients. In the case of endobronchial, one-lung ventilation, the endotracheal tube can be positioned in the right or left main stem bronchus until the fistula is ligated surgically, then withdrawn into the trachea for the remainder of the procedure. Given the difficulty of ventilating the right upper lobe (even with careful positioning of the Murphy eye), left mainstem intubation is often preferred. One lung ventilation offers the surgical advantage for less movement of the operative field for thoracoscopic or open repairs. However, a patient with preexisting lung disease from chronic aspiration or respiratory distress syndrome of prematurity may not tolerate single lung ventilation. When possible, for small/proximal tracheal fistulas, the endotracheal tube can be advanced just distal to the fistula, allowing for ventilation of both lungs. An endotracheal tube without a Murphy eye and positioning of the bevel to face anteriorly will maximize occlusion of the fistula [27]. Extreme care should be taken to prevent migration of the endotracheal tube into or past the fistula, which can cause life-threatening desaturation [28]. Other endotracheal tube positions can be tailored to the nature of the particular patient's anatomy. For example, one case report described placing a standard oral endotracheal tube placed just distal to the fistula, with the tip of the tube in the right bronchus, and the Murphy eye ventilating the left lung [29]. Intraoperative bronchoscopy is extremely helpful in positioning, but if unavailable in LMIC, the tip of the tracheal tube can be positioned just above the carina by auscultation of diminished breath sounds over the left axilla as the tube is advanced in to the right mainstem bronchus, after which the tube is retracted until breath sounds are once again increased over the left chest [30].

Occlusion of the fistula with a Fogarty arterial embolectomy catheter is a preferred method for securing the airway in patients with large, distal fistulas and complex medical history [31]. Placement of the catheter is performed after induction of general anesthesia, but prior to intubation, under bronchoscopic guidance. The Fogarty catheter is visualized and advanced into the TEF, and then the trachea is intubated in the standard fashion with an oral endotracheal tube [32] This technique virtually eliminates intraoperative desaturation episodes while thoracoscopy or thoracotomy is performed to ligate the fistula [31].

Finally, the role of fiberoptic tracheoscopy cannot be understated. Appropriately sized pediatric fiberoptic bronchoscopes, in the hands of a skilled bronchoscopist, can provide tremendous information about tracheobronchial anatomy, TEF location and size, and endotracheal tube positioning. It can also assist surgeons in ligation of the fistula using surgical transillumination, and assessment of bleeding, secretions, and residual tracheomalacia intraoperatively [32, 33].

Post-operative ventilation for patient's requiring continued ventilation care is also an area of interest. The literature is lacking in regard to optimal method for providing ventilator support for TEF/EA managed with open repair. For centers to be performing open or thoracoscopic repair, the center must have methods of effectively ventilating these babies. One study recommends for non-reversal of anesthesia and stepwise withdrawal of respiratory support that has shown to have a beneficial effect [2].

Based on these considerations, and review of case series, a possible approach to management of the airway is as follows [34]:

1. Induce general anesthesia by inhalation or intravenously (taking care to maintain spontaneous respiration)
2. Demonstrate adequate face mask ventilation with low inflation pressures, without causing gastric distention (Optional—induce muscle relaxation and again demonstrate adequate mask ventilation)
3. Rigid bronchoscopy (if desired by the surgical team) to further characterize the airway, main fistula, presence of secondary fistulas, or other anomalies such as vascular rings. A Fogarty catheter can be placed during this step to occlude the fistula.
4. Intubation either beside the Fogarty catheter, or below the fistula as described earlier

After induction of general anesthesia and intubation, these cases warrant a second venous line and an arterial line [35]. Blood loss is usually minimal during this procedure.

Most infants return to the intensive care unit postoperatively intubated and sedated. Although many patients undergoing a straightforward repair can be extubated within 24 h, one should not adopt a cavalier approach about early extubation because of the dangers of a traumatic intubation in the setting of airway edema and fresh tracheal suture lines. Post-operative analgesia may be administered by intravenous opioids, epidural catheter, subcutaneous wound catheter, or local infiltration. There is no evidence that one technique provides and benefit over the others, and preference is dictated by local expertise, experience, and access to resources [36].

Conclusion

Significant challenges for repair of tracheoesophageal fistula still exist in the wide spectrum of 'the developing world'. The authors suggest a realistic assessment of the capabilities of each individual institution including the patient referred, instrumentation available, surgeon experience, anesthetic capabilities and comfortability, post-operative intensive care, and subspecialty support. If the context allows, a gradual

introduction of advanced minimally invasive repair can be attempted in the developing world.

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