Thoracoscopic primary repair of esophageal atresia and tracheoesophageal fistula: Our early experience

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ABSTRACT

Background: Thoracoscopic repair of esophageal atresia (EA) and tracheoesophageal fistula (TEF) has been practiced more frequently in the past two decades. This study focuses on assessing our early experience with implementing this minimally invasive technique in neonates.

Methods: Neonates with EA/TEF who were admitted to the NICU at Ain Shams University hospitals during the period from 2021 to 2022 were enrolled in the study. Thoracoscopic primary repair was performed for neonates with a body weight exceeding 2 kg. Neonates with associated major cardiac, pulmonary, and neurological anomalies and long gap atresia were excluded from the study.

Results: A total of 38 neonates presented with EA/TEF during the study period. Out of these neonates, 12 cases underwent thoracoscopic primary repair. The thoracoscopic approach was used to complete all necessary repairs, and no intraoperative complications occurred. The mean operating time was 125 minutes, ranging from 105 to 150 minutes. No evidence of leakage was found in any case, as determined by both clinical and radiological contrast swallow examinations. The mean ventilation time was 7.2 days, and the mean hospital stay was 21 days. There were four deaths attributed to complications related to barotrauma and sepsis.

Conclusion: Thoracoscopic repair of EA can be performed safely, which yields favorable functional and cosmetic outcomes. However, it remains a challenging procedure that requires a long learning curve.

INTRODUCTION

The history of EA dates back to William Durston's description in 1670 [1]. However, the most significant contribution occurred in 1939, when a neonate survived the procedure [2,3]. In 1941, Dr. Cameron Haight performed the first successful primary repair by making a left thoracotomy [4]. The approach to EA repair has evolved over the years, marked by significant milestones such as the first thoracoscopic repair in 1999 by Lobe et al. Another notable advancement was the first thoracoscopic repair of EA with TEF in 2000 by Rothenberg [5,6]. Several studies, such as the 2005 review by Holcomb et al. and the 2015 multi-institutional study by Okuyama et al., have examined the outcomes of thoracoscopic and open repair procedures, finding comparable results [7,8]. Elbarbary et al. (2020) highlighted the importance of thoracoscopy and recommended that

pediatric surgeons acquire the necessary expertise to perform this procedure [9].

This study aims to assess the feasibility, safety, and short-term outcomes of thoracoscopic repair of EA and TEF in our center. Additionally, it seeks to contribute to the existing knowledge by reporting the outcomes of our early experience with this surgical technique. This study will provide a learning pathway for beginners to gain expertise in thoracoscopic EA repair.

METHODS

From January 2021 to December 2022, Neonates presented with EA/TEF at Ain Shams University hospitals were enrolled in this study. Inclusion criteria was type C esophageal atresia amenable for primary anastomosis. Exclusion criteria were patients with a birth weight < 2 kg, long gap EA, and patients

with major cardiac, pulmonary, or neurological anomalies. The study was approved by the ethical committee of our faculty (IRB no. 00006379). Informed written consent was taken from the patient's guardian illustrating the benefit and possible complications of the procedure. The probability of conversion to the open technique was explained to the parents as this was an early experience.

The diagnosis was established using a plain chest X-ray with the insertion of a nasogastric tube 10 Fr to detect the level of the upper pouch. A comprehensive preoperative assessment was conducted, including laboratory investigations, a pelvis-abdominal ultrasound, and an echocardiogram. The purpose of these tests was to rule out any associated anomalies and to determine the site of the aortic arch.

The operative technique

General endotracheal anesthesia was performed, and efforts were made to maintain modest peak pressures. The patient was positioned in a prone position at the edge of the table, with the chest and waist elevated, making room for the abdomen to allow for breathing. In addition, the right arm was elevated towards the patient's head (Fig. 1). The surgeon and the assistant stood on the same side of the table, and the monitor was placed on the other side. A Nelaton catheter was inserted through the nostril at the upper esophageal pouch to facilitate its identification during the thoracoscopy.

Figure 1: Patient's position and the port sites.

Three ports were used for right thoracoscopy: a 5 mm port for optical trocar and two 3 mm ports for instruments. The first optical port was placed below the angle of the scapula, and a 30º lens was used. The right-hand port usually landed high in the axilla. The left instrument was placed 2 cm medial to the optical trocar. A low flow, low pressure of CO_2 was administered to collapse the right lung (4 mmHg, 1 L/min).

The major landmark identified was the azygos vein where the fistula lies below it. We kept the azygos vein intact in all cases. The pleura below the azygos vein was incised to identify the lower pouch with the vagus nerve overlying it. The lower pouch fistula was then gently grasped with the nontraumatic bowel grasper and dissected, preserving the vagus nerve. The fistula was precisely dissected to its junction with the trachea. The fistula was then ligated with a 4-0 polyglactin suture (Vicryl ®).

Afterward, the upper pouch was identified and carefully dissected from the trachea. The feasibility of primary anastomosis was evaluated by examining the gap between the upper and lower pouches. The upper pouch was then mobilized all around to gain length for tension-free anastomosis. Once mobilized, the distal tip of the upper pouch was incised. At this point, the fistula was divided; the lower pouch was sharply cut below the tied fistula, and the two ends were now ready for anastomosis.

Anastomosis was performed using intracorporeal sliding square tumble knots. After placing the first knot, the posterior row sutures were done on both sides of the midline suture, ensuring mucosa was included, using 5-0 absorbable polyglactin sutures (Vicryl ®) in a single interrupted fashion. Once the posterior wall was completed, an 8 fr. nasogastric tube was passed through the anastomosis. Subsequently, the anterior wall anastomosis was completed, and a 12 Fr chest drain was placed under vision with the tip placed near the anastomosis. The port sites were sealed and infiltrated with local anesthesia (Fig. 2).
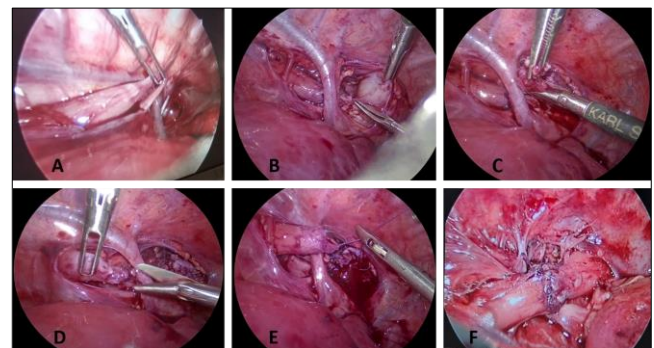

Figure 2: the operative procedure: A; Lower pouch dissection with the fistula elevated showing the junction with the trachea. B; Identification of the upper pouch and its dissection from the overlying fascia. C; Incision of the tip of the upper pouch. D; The lower pouch is incised below the tied fistula. E; Tumble sliding intracorporeal knot for the first stitch at the middle of the posterior wall. F; After completion of the anastomosis.

Postoperatively, the patients were transferred to the neonatal intensive care unit and kept ventilated till respiratory status allows safe extubation. We keep them on antireflux position with administration of antireflux medications mainly in the form of proton pump inhibitors. Single antibiotics and adequate

analgesic were given. Usually, we started nasogastric feeding on the next day if the general and respiratory conditions permit.

RESULTS

During the study period, 38 neonates presented with EA/TEF. Sixteen cases underwent thoracoscopic intervention, with 12 of them undergoing thoracoscopic primary repair. Four cases were excluded due to long-gap atresia; three of these underwent a traction procedure, and one required esophagostomy and gastrostomy.

A total of 12 neonates (eight males and four females) underwent thoracoscopic primary repair. In the preoperative X-ray, the level of the upper esophageal pouch was at T2 in 2 cases, T3 in 7 cases, and at T4 in 3 cases. That was also confirmed intraoperative to be a short gap EA amenable for tension free primary anastomosis. Their gestational age ranged from 35 to 40 weeks (median = 37), their birth weight ranged from 2.1 to 3.5 kg (median =2.8), median age at presentation was two days (ranged from 1 to 8 days), and median age at surgery was three days (ranging from 2 to 12 days).

Preoperative Echocardiography showed mild defects in 10 cases (62.5%). These included PFO (43%), PDA (31%), mild to moderate TR (43%) with mild pulmonary hypertension, and ASD (12.5%). All patients had left-sided aortic arch. No major cardiac anomalies were reported in this series.

Four patients required mechanical ventilation prior to the surgery. Three of these cases were a result of delayed referral to our center on days 6, 7, and 8. The fourth patient experienced worsening respiratory distress. Preoperative ultrasound showed lung consolidation and turbid pleural effusion, and thoracoscopy revealed some purulent secretions around the lung with mild adhesions, although presented on day 2 and operated on day 4.

The mean operative time for primary repair was 125 ± 18 minutes (ranging from 105 to 150 minutes) (median = 120) (Fig. 3). Time needed for dissection and ligation of the fistula was 10 to 15 minutes. No intraoperative complications were encountered during this series. All cases were completed thoracoscopically with no conversion to open thoracotomy.

All cases were transferred from the OR to the NICU on mechanical ventilation with an endotracheal tube. Total mechanical ventilation time ranged from 4 to 14 days (median = 8). In addition, nine patients (60%) required cardiac support postoperatively.

Barotrauma was a significant complication encountered in this series, with no evidence of anastomotic leak. Nine cases (56.25%) had

barotrauma (bilateral in 4 cases), which needed relief using another chest tube other than the drain kept during the surgery.

Nasogastric tube feeding was started early on day 1 to day 3 postoperatively according to the patient's general condition.
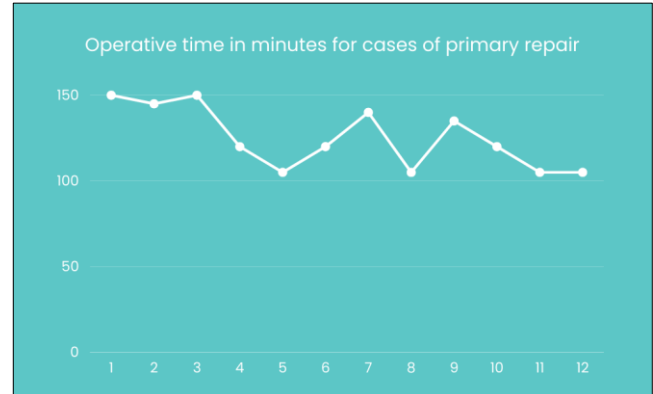

Figure 3: Line graph showing operative time in minutes along the progress of cases.

On day 7, a contrast swallow study was performed on eight cases. The study showed a well-formed anastomosis with a good diameter, and the contrast passed freely to the stomach without any leakage. Contrast was delayed in 3 cases due to difficult transportation as the patients were on mechanical ventilation and performed at 10, 15, and 16 days postoperative, revealing no leak. The contrast was not done in one case; the baby experienced severe sepsis due to bilateral pneumonia, and he was on high-frequency ventilation. However, the patient died on day 15 postoperatively without clinical or radiological (plain X-ray) evidence of leakage. The median total hospital stay was 23 days ranging from 16 to 30 days (mean 21 ± 5 days).

In this series, four cases died. Mortality can be attributed to the following morbidities: barotrauma in 4 cases (100 % of mortality cases), pneumonia in 3 cases (75 % of mortality cases), sepsis in 4 cases (100 % of mortality cases), and pulmonary hypertension in 2 cases (50% of mortality cases).

After a 6-month follow-up, the eight patients who survived gained appropriate weight gain. One patient exhibited radiological reflux during the contrast study conducted six months later despite the absence of significant symptoms. Antireflux management was prescribed for 6 months and we planned to repeat the contrast study at age of 1 year, however, the parents refused to repeat the contrast as their baby was doing well without any clinical symptom.

One patient presented with a mild esophageal stricture, which required one session of endoscopic esophageal dilatation at the age of 10 months. After this session the patient was followed up for another

18 months where he was dysphagia free all through this period. Moreover, cosmetic outcomes were satisfactory, with no disfigurements and without the musculoskeletal sequelae associated with the classic open thoracotomy.

DISCUSSION

Over the past two decades, the thoracoscopic repair of EA/TEF has become more widely practiced to improve results by avoiding thoracotomy and its subsequent morbidities [10]. Several studies report that thoracoscopic repair is effective and safe, yielding comparable results to open repair. Additionally, thoracoscopic repair offers improved visualization of posterior mediastinal structures and eliminates the musculoskeletal complications associated with thoracotomy [11,12].

Despite its advantages, minimally invasive EA/TEF repair remains technically challenging, and anastomosis suturing may be a significant barrier. That's why a few reports have indicated that patients with LBW should be excluded from the indications. These studies considered LBW less than 2 kg [13,14] or below 2.5 kg [15, 16]. In our study, we excluded patients with birth weights below 2 kg. The patients' birth weight ranged from 2.1 to 3.5 kg (median =2.8).

Our study's median age at operation was three days (ranging from 2-12). In the study of Yang and his colleagues,[17] the median age at the time of operation was 4 (with a range of 3-7) days. In the study conducted by Rozeik et al. [12], the median age at the time of the operation was six days, ranging from 2 to 26 days. In our experience, we prioritize early intervention to minimize the risk of developing aspiration pneumonia.

EA is frequently associated with other congenital anomalies, particularly those with the VACTERL association [18]. The most frequently reported associated anomalies with EA/TEF are cardiovascular anomalies, with an incidence rate of 67%, followed by spine, gastrointestinal, genitourinary, and limb anomalies [19]. The present study showed an overall prevalence of cardiac anomalies of 62.5%, mostly patent foramen ovale (43%), mild to moderate tricuspid regurge (43%) with mild pulmonary hypertension, and patent ductus arteriosus (31%). Although there were a significant number of cardiac anomalies in our study, none impacted the patients' hemodynamic status or cardiac functions.

Preoperative ventilator dependence is an additional prognostic factor independent of the associated anomalies. These factors may have a more significant impact on survival than body weight [20]. In our study, 25% of cases needed mechanical ventilation before operation. Additionally, 50% of these cases were subsequently reported as mortality. However, this percentage is inadequate to establish an association between preoperative ventilation and postoperative mortality.

The operative time of thoracoscopic repair for EA varied among different case series. Anatomical factors, such as small body weight, tiny distal esophagus, and long gaps, account for the longer operative time. The operative time for thoracoscopic repair of EA can be influenced by the significant learning curve associated with this procedure, as well as the experience of the surgical team. The literature review conducted by Okuyama et al. [10] reported a median operative time ranging from 100 to 230 min.

The mean operative time in the study of Elbarbary et al.[9] which included 41 thoracoscopic primary repair cases was 108.3 minutes (range 80-122). In a study conducted by Shirota et al. [14], it was found that the average duration of surgery in 26 cases of primary repair was 149 minutes, with a range of 73 to 245 minutes. A retrospective study was conducted at Beijing Children's Hospital, involving 190 patients who underwent surgery for EA over 13 years. Out of these patients, 62 cases were operated on using thoracoscopy, with a median operative time of 125.5 minutes (range 90.0-206.3) [17].

The present study's mean operative time for primary repair was 125 minutes (range 105-150). Despite our early experience, our mean operative time did not differ from what was reported in the previous series. We attribute this to our experience in other different laparoscopic operations, so we think that experienced pediatric laparoscopic surgeons can use thoracoscopy in EA/TEF repair safely and effectively. The operative time also decreased over the study period.

Shirota and his colleagues reported conversion to open surgery in 5–15% of their cases, and this was attributed to the inability to perform anastomosis because of strong tension, bleeding, and poor visualization.[14] However, all procedures were completed in our case series without conversion to open surgery. We think that thoracoscopy provided us with good visualization and allowed us to adequately dissect both the upper and lower pouches without needing conversion to complete the procedure. We think that conversion would be needed if we faced a major vessel injury with uncontrollable bleeding, which was not encountered in this series. Conversion and intraoperative complications may have also been avoided by excluding cases with fragile tissues weighing < 2 Kg.

The most common complications after repair of EA/TEF are related to anastomosis. The observed leakage rate varies from as low as 2.5% to as high as 33.5% [12]. The literature review conducted by Okuyama et al. [10] reported an anastomotic leakage rate ranging from 0 to 20%. A variety of factors can

contribute to the leakage, such as excessive mobilization of the distal end of the esophagus, esophageal injury, poor suturing technique, anastomotic tension, the use of inappropriate sutures, associated ischemia, a long gap length or sepsis [20].

In the current study, no instances of leakage were encountered. As this work was an early experience to assess the feasibility and safety of the technique in our hands, we selected the cases who are amenable to tension-free primary anastomosis excluding long-gap cases. So, the absence of leakage in this series could be attributed to the selection of simple cases to start with rather than complicated cases where the incidence of leakage is higher. Nevertheless, barotrauma was one of the significant complications encountered during our study. This was a major risk factor for mortality due to sepsis and deterioration of the general condition. All these patients needed intervention for relief of the pneumothorax by intercostal tube insertion. In the study of Yang et al. [17], they had a rate of pneumothorax in the thoracoscopic group of 41%. However, only 40% of these cases were observed, and 60% needed drainage.

In the study of Shirota et al. [14], the prevalence of GERD was approximately 70%. The issue was resolved through a combination of conservative measures and careful monitoring. An anastomotic stricture was detected in the early phase in around 10% of the patients. However, it should be noted that the stricture can also manifest more than one year after the surgery. According to Galazka et al. [20], 15% of patients had such severe GERD that they required an anti-reflux procedure called Nissen fundoplication. During our study, we observed one instance of gastroesophageal reflux that did not require surgical intervention and another case of anastomotic stenosis that required one session of endoscopic dilatation.

The overall mortality rate in the present study was 33.3 %, with sepsis being the most prevalent cause. Sepsis is a prominent factor in the elevated mortality rate of neonates in developing countries. This finding may be attributed to the poor respiratory health of the patients and the need for prolonged postoperative ventilation [9]. Several studies have identified sepsis as the leading cause of mortality [21,22,23]. The literature review by Okuyama et al. [10] found that the mortality rate was reported to range from 0 to 21%. The high mortality rate in our series can be attributed to the early experience in such management strategy, as previous research has indicated a substantial learning curve in the first 10-20 cases, with better outcomes anticipated in subsequent cases [10,24,25].

The limitations of our study included small cohort size, limited use of thoracoscopy in our center during the COVID-19 era, lack of comparative groups with the conventional open repair, which is the established technique in our center, and absence of long-term follow-up.

Despite the great advantage of the thoracoscopic approach for EA/TEF repair, it is a challenging technique. Surgeons should have the ability to operate precisely and safely within so limited space in a very critical neonate. For us to be able to do that, we selected our team from the most expert minimal invasive pediatric surgeons in our department. Thereafter, we attended many minimally invasive workshops focusing on thoracoscopic EA/TEF repair including hands-on training (wet animal lab), especially for training on intra-corporeal suturing. And then for a few cases; we did only the part of fistula ligation and upper pouch dissection thoracoscopically and completed the anastomosis with the open approach. And at a point where we felt so familiar with the field, we started this work with complete thoracoscopic repair.

CONCLUSION

Our findings indicate that thoracoscopic repair of EA can be conducted safely, resulting in favorable functional and cosmetic outcomes. Additionally, this approach offers all the advantages associated with minimally invasive surgeries. Nevertheless, this procedure is still complex and requires a significant amount of time to learn. It should only be done in pediatric centers with the necessary equipment and a team of experts in neonatology, pediatric anesthesia, and pediatric surgery who are experienced in performing minimally invasive surgery. This is important to ensure the highest level of care in hemodynamic and respiratory monitoring.

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