



Thoracoscopic versus conventional open repair of tracheoesophageal fistula in neonates: A short-term comparative study

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ABSTRACT

Purpose: Esophageal atresia with or without a tracheo-esophageal fistula is a challenging anomaly in neonates. Thoracoscopic repair is gaining popularity now in pediatric surgery community. The present study aims at comparing the short term outcomes of thoracoscopy versus classic thoracotomy for repair of such conditions.

Methods: Thirty neonates with tracheoesophageal fistula were randomly divided into two equal groups ($n=15$) after excluding patients with birth weight <2000g, multiple associated anomalies and cardiorespiratory instability. One group had conventional open repair while the other had thoracoscopic repair. Demographic data, intraoperative result and post-operative findings were recorded and compared between both groups.

Results: Both groups showed similar results regarding demographic and patients' characteristics. Thoracoscopic repair had relatively longer, yet non-significant operative time but with highly significant difference in preserving azygos vein. There was low conversion rate with thoracoscopy (6.66%). Open repair resulted in a longer hospital stay (11.73 ± 5.68 vs 9.2 ± 2.95). Complication rate was comparable in both groups; however, thoracoscopy was associated with better cosmetic results as reported by parents and surgeons ($p=0.00$).

Conclusion: Compared to thoracotomy, thoracoscopic repair offers a less invasive, effective and safe technique with similar short term outcomes, but with superior cosmetic results and better ability to spare azygos vein.

Type of Study: Therapeutic/Treatment study

Level of Evidence: Level II

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Esophageal atresia (EA) with or without a tracheo-esophageal fistula (TEF) is a challenging anomaly in neonates. Traditionally these patients have presented shortly after birth because of an inability to pass an orogastric tube, respiratory distress, or an inability to tolerate feedings. The condition may be associated with other major congenital anomalies (VACTERL syndrome), or may be an isolated defect [1].

Conventionally, open, right-sided, muscle-sparing thoracotomy is the standard approach for repair of EA/TEF in many surgical centers [2]. Several reports have shown the effectiveness and safety of thoracoscopic repair (TR) [3,4].

Allal et al. [5] confirmed the reliability of TR, and affirmed a reduced requirement for analgesia with comparable outcomes. Yamoto et al. [6] demonstrated that the thoracoscopic approach was favorable and safe for EA/TEF repair in carefully selected patients while Koga et al. [7] confirmed that TR is less traumatic than conventional open repair (COR) as direct retraction of the lung is unnecessary, which results in lower impact to the respiratory tract and a smoother recovery.

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A meta-analysis carried out in 2012 concluded that there were no significant differences between TR and COR with respect to leaks and strictures [8]. This was confirmed by another study in 2016 which concluded that, compared with COR, TR is associated with a similar complication rates of leaks and strictures, and longer operative time [4].

The present work aims at comparing the short term outcome of thoracoscopic versus conventional open repair of tracheoesophageal fistula with esophageal atresia in neonates.

1. Patients and Methods

After obtaining Institution Review board approval (#3638/23-4-2017), thirty neonates admitted to Zagazig University Hospitals and Cairo University Children Hospital with esophageal atresia and tracheoesophageal fistula in the period between July 2017 and January 2019 were enrolled in the study (after exclusion of patients with long gap or pure esophageal atresia in whom primary anastomosis cannot be done, patients with birth weight <2000 g due to increased surgical and anesthetic difficulties facing the still developing learning curve in neonatal thoracoscopic surgery in our center, patients with severe cardiac malformations e.g., single ventricle and chromosomal aberrations

e.g., trisomy 18 and trisomy 13 or more than one major associated malformation such as duodenal or anal atresia and unstable patients regarding hemodynamics and cardiorespiratory state). Patients were divided into two equal groups (n=15) using a computer-generated randomization table assigning patients randomly into either COR group or TR group. The study was non-blinded but both groups were operated by the same surgeons, managed and treated equally regarding the post-operative care, frequency of follow-up and management of complications.

1.1. Pre-operative

All patients had preoperative evaluation including physical examination, plain chest x-ray with 8F nasogastric tube inserted with no pressure placed on it for leveling the upper pouch and detection of type of atresia and measuring the gap length, using the lowest point of the coiled NGT as a land mark for the upper pouch and a point just above the carina (T4) as a mark for the fistula and lower pouch (>3 vertebral bodies was considered long and excluded). The gap length was estimated using the same technique by the same surgeons for all cases. Contrast swallow was used cautiously when indicated. All patients had ECHO done preoperatively for detection of cardiac anomalies and the locating the aortic arch. Ultrasound was done for detection of renal anomalies. Patients were admitted to surgical NICU and kept NPO with continuous suction of the upper pouch and nursed in a head-up position to minimize aspiration. Antibiotics and IV fluids were initiated and patients prepared for surgery. Informed consent was obtained from all patients' parents.

1.2. Surgical technique

1.2.1. COR group

After general anesthesia was started, patient was positioned in a Lt. lateral position. A classic right lateral thoracotomy incision one finger breadth below the tip of scapula (Fig. 1) with partial dissection of the latissimus dorsi as well as the serratus muscle was performed. An intra- or extra-pleural approach to the mediastinum was used. The azygos vein was spared whenever possible or ligated according to need. After identification and ligation of the fistula, the lower pouch and kept in place and was not divided until identification and mobilization of the proximal esophagus was done (Fig. 2). With the two ends mobilized, the anastomosis is performed using 5/0 polygalactin sutures in a single interrupted fashion. The posterior wall was anastomosed first and then the NG tube was passed under vision into the lower pouch and stomach. The anterior wall was then completed. Chest drain was placed in all cases. Closure of the thoracotomy was carried out in anatomical layers.

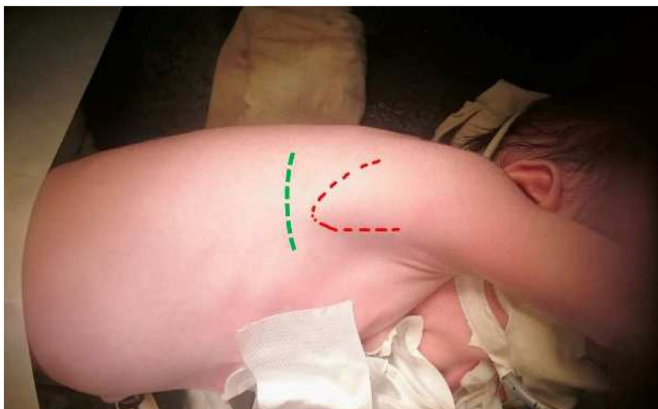


Fig. 1. Patient positioning and site of incision (green line) in COR.

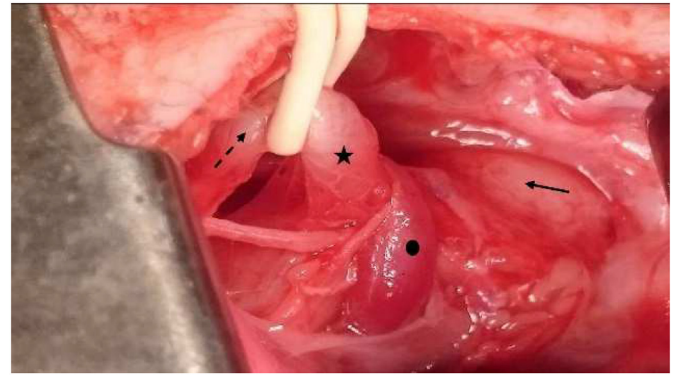


Fig. 2. Identification of the fistula (star), lower pouch (dashed arrow), upper pouch (arrow) and the azygos vein is seen intact crossing over the fistula (circle).

1.2.2. TR group

The patient was placed in a prone position with the right side slightly elevated and the patient is placed as near as possible to the edge of the operating table to allow free movement of the instruments. The initial port (5mm) was placed one finger breadth below the lower angle of the scapula. A 5-mm 30° angled scope was used through this port for visualization. Two 3mm working ports were used. The first port is placed in the same costal space as the camera port 3 cm from midline. The second port is placed as high as possible in the axilla. Low pressure of CO₂ (4–6 mmHg, 1 L/min) was used to collapse the right lung and create working space aided by the natural gravity pulling the right lung down away from the field.

Once the chest has been insufflated and the lung collapsed, the fistula was identified. With the azygos vein spared whenever possible, the lower esophageal segment was identified and dissected bluntly to preserve the aortic branches. After complete mobilization of the lower segment, the fistula can be safely sutured and divided using a transfixing suture (Fig. 3). The anesthesiologist places pressure on the NG tube to help identify the upper pouch. The pleura overlying the pouch was dissected and the pouch was mobilized with blunt dissection. Once adequate mobilization is achieved, the distal tip of the pouch was opened. With the two ends mobilized the anastomosis is performed using single layer 5/0 polygalactin interrupted sliding tumble square knots. The posterior wall was anastomosed first and then a 6F or 8F NG tube was passed into the lower pouch and into the stomach. The anterior wall was then completed over the NG tube. After the anastomosis was completed, a chest tube was placed when needed through the lower trocar site and the tip is placed near the anastomosis and port sites were closed.

1.3. Post-operative

Post-operative, the patient was transferred back to the NICU, where he was weaned of mechanical ventilation, nursed in a head-flexed position. Routine post-operative esophagograms were obtained on the sixth post-operative day and oral feedings were started after ensuring no evident leakage is present. Chest drain was removed and feeding increased, patients were discharged when tolerating full feeding with no fever and good general condition.

Data were collected regarding gender, gestational age, birth weight, age at presentation and at operation and associated anomalies. Intraoperative data were collected including operative time, completion or conversion, site of fistula, division of azygos vein or not, measured gap length and intra-operative complications. Time to extubation, time to first oral feeding, early post-operative complications as anastomotic leak, stricture, lung complications, wound infection, duration of hospital stay and mortality were recorded. Patients were followed-up in outpatient clinic for 6 months for detection of delayed complications as esophageal stricture. Evaluation of the scar was done using modified

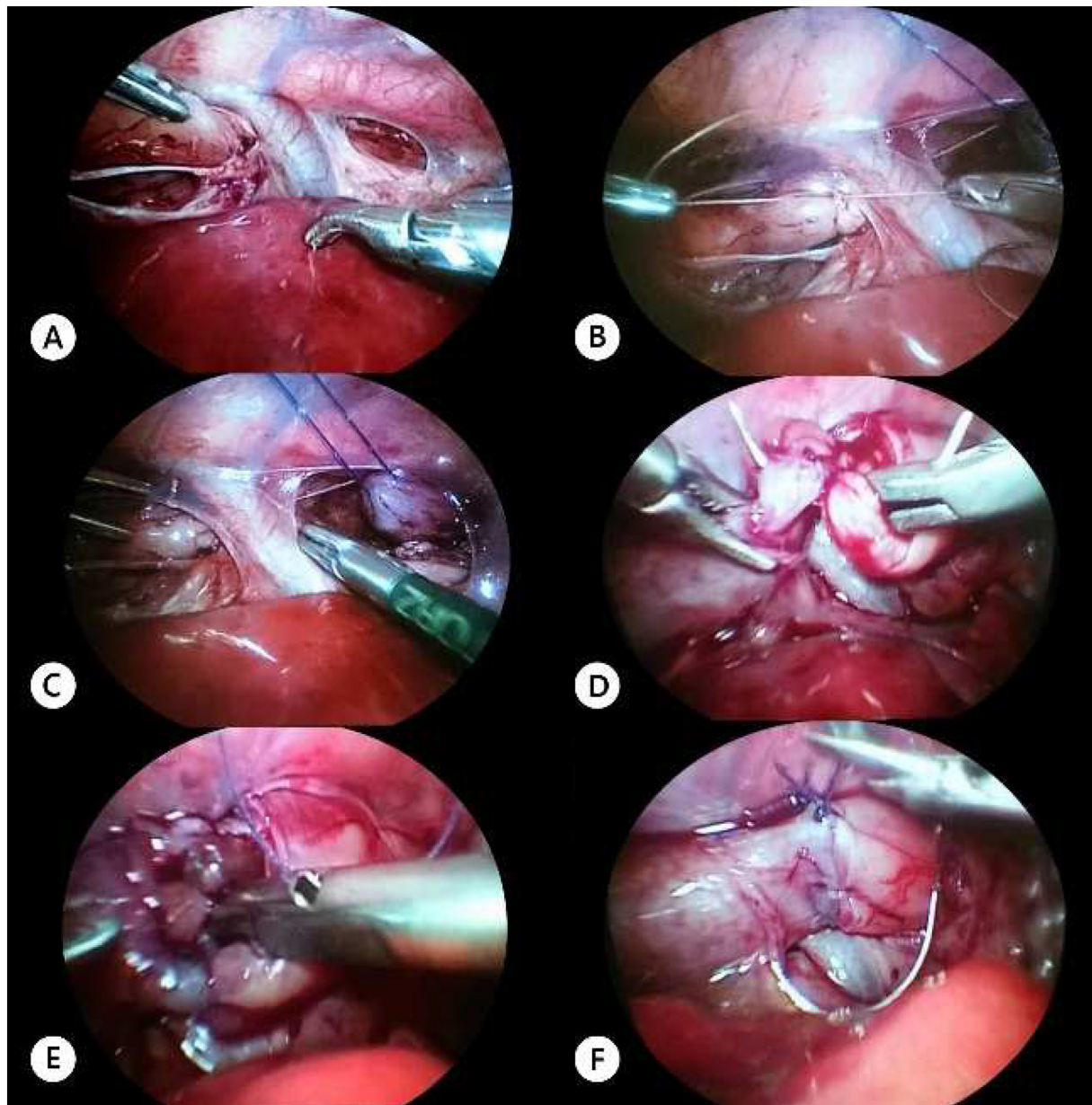


Fig. 3. Thoroscopic repair of EA/TEF; A) Initial view with identification and dissection of lower pouch, B) Identification and ligation of the fistula, C) Dissection of the upper pouch, D) Suturing the posterior wall, E) Passing a 6F nasogastric tube through anastomosis into the stomach, F) Final sutures of the anterior wall and completed anastomosis.

Patient and Observer Scar Assessment Scale (POSAS) [9] which takes into account both the patient and provider perspective, including both a patient scar assessment scale (P-SAS) and an observer scar assessment scale (O-SAS).

1.4. Statistical analysis

The collected data were computerized and statistically analyzed using SPSS program (statistical package for social science) version 18.0. Qualitative data were represented as frequencies and relative percentages and analyzed using Chi-square test, while quantitative data were expressed as mean \pm SD (Standard deviation). Independent t-test was used for quantitative variables in normally distributed data while Mann Whitney (MW) test was used for quantitative variables in not normally distributed data. For all analyses, a p value of ≤ 0.05 was considered significant.

2. Results

Regarding demographic variables and patients' characteristics (**Table 1**), there was male predominance in both groups (1.5:1, 1.14:1 respectively) but with no statistical significance. Gestational age was comparable (36.25 ± 2.21 vs 37.06 ± 1.48 weeks). There was no statistical significance regarding birth weight, age at presentation, age at operation. Regarding cardiac anomalies, 80% of COR group patients had 23 cardiac anomalies (some patients had more than one anomaly), with PDA and ASD being the commonest anomalies in this group. Meanwhile, 86.6% of TR group patients demonstrated 25 cardiac anomalies with PFO and PDA being the commonest lesions respectively. None of the associated cardiac anomalies showed statistically significant difference between the two groups.

Anorectal malformations (ARM) was found in 2 patients in COR group, however, no patients in TR group suffered from ARM. Renal anomalies was the commonest associated anomalies in COR group

Table 1
Demographic data and patients' characteristics in both groups.

Variable	COR group (n = 15)	TR group (n = 15)	Test	p-Value
Gender				
Male	9 (60%)	8 (53.33%)	χ^2	0.71
Female	6 (40%)	7 (46.66%)		
Male:Female Ratio	1.5:1	1.14:1		
Gestational age (wk)				
Mean + SD (Range)	36.26±2.21 (32–40)	37.06±1.48 (34–39)	t-Test	0.26
Birth weight (g)				
Mean + SD (Range)	3006±413.05 (2550–3750)	2936.66±306.4 (2400–3450)	t-Test	0.61
Age at presentation (d)				
Mean + SD Median (range)	3.53±3.22 2 (1–12)	5.13±5.79 3 (1–24)	MW	0.36
Associated anomalies				
Cardiac	12 (80%)	13 (86.66%)	χ^2	0.62
Anorectal	2 (13.33%)	0 (0%)		
Renal	3 (20%)	1 (6.66%)		
Limb	0 (0%)	0 (0%)		0.28
Vertebral	0 (0%)	0 (0%)		—
Down syndrome	1 (6.66%)	1 (6.66%)		1.00
Undescended testis	1 (6.66%)	0 (0%)		0.31

SD: Standard Deviation; **P-SAS:** Patient Scar Assessment Scale; **O-SAS:** Observer Scar Assessment Scale; **COR:** Conventional Open Repair; **TR:** Thoracoscopic Repair; **MW:** Mann-Whitney test.

including renal agenesis, grade I nephropathy and hydronephrosis. Down syndrome was found equally in only 1 patient in both group, while only one patient in COR group had a unilateral undescended testis. None of these associated anomalies had statistical significance compared to the other group.

Regarding operative results (Table 2), the mean operative time was comparable in both groups (94.13±16.6 vs 104.66±17.47 respectively) with no significance of the operative technique on the surgery time. Meanwhile, azygos vein was significantly spared in TR group (100%), unlike COR group, where sparing the azygos vein was possible in only five patients (33.33%). On the other hand, it was ligated and divided in the rest 10 patients (66.66%) to allow better visualization of the mediastinal structures.

Table 2
Intraoperative findings in both groups.

Variable	COR Group (n = 15)	TR Group (n = 15)	Test	P-value
Age at operation (d)				
Mean + SD Median (Range)	6.33±3.71 5 (2–15)	7.2±5.90 6 (2–26)	MW	0.63
Operative time (min)				
Mean + SD (Range)	94.13±16.6 (70–120)	104.66±17.47 (80–140)	t-Test	0.10
Azygos vein status				
Spared	5 (33.33%)	15 (100%)	χ^2	0.00** (HS)
Ligated and divided	10 (66.66%)	0 (0%)		
Site of the fistula				
Lower Pouch	15 (100%)	15 (100%)	χ^2	—
Upper Pouch	0 (0%)	0 (0%)		
Both	0 (0%)	0 (0%)		
Gap length (vertebrae)				
Mean + SD (Range)	2.27±0.54 (1.2–3)	2.34±0.63 (1.2–3)	t-Test	0.65
Intraoperative comp.				
Number	1 (6.66%)	1 (6.66%)	χ^2	1.00
Conversion	--	1 (6.66%)		

SD: Standard Deviation; **P-SAS:** Patient Scar Assessment Scale; **O-SAS:** Observer Scar Assessment Scale; **COR:** Conventional Open Repair; **TR:** Thoracoscopic Repair; **MW:** Mann-Whitney test.

** HS: Highly significant (p<0.01)

TEF was Gross type C (fistula to the lower pouch) in 100% of patients in both groups. The gap length between upper and lower pouch was measured in vertebral length with no statistical significance found (2.27±0.54 vs 2.34±0.63). Intraoperative complications occurred equally in both groups. In COR group, one patient (6.66%) had an intraoperative event in the form of injury to the upper trachea while dissecting the upper pouch from the adherent trachea. Repair of the trachea was done with interrupted sutures and the patient recovered well. On the other hand, in TR group one patient (6.66%) had an iatrogenic injury to the left main bronchus during dividing the fistula, the procedure was converted to COR and repair of the bronchus was done with interrupted sutures and procedure completed in open fashion.

No statistical significance was found between the two groups regarding days to extubation, days to first oral feeding and total duration of hospitalization.

In COR group, two patients (13.33%) were reintubated, one at 12th POD because of respiratory failure associated with renal impairment and sepsis, and unfortunately he died at 14th POD, and the other was on the fourth POD and died on the fifth POD due to deterioration of pre-existing aspiration pneumonia-related sepsis. On the other hand, in TR group, one patient (6.66%) required reintubation at fifth POD due to Rt. Sided pneumonia with tachypnea and acidosis.

Wound infection and skin dehiscence was the commonest complication in COR group (20%), however, it didn't occur at all in TR group (0%). Anastomotic leakage occurred equally in both groups (13.33%), leakage was managed conservatively in both cases in COR group, while in TR group, one patient was successfully managed conservatively and one patient needed gastrostomy and esophagostomy. Stricture occurred in 2 patients in COR group (13.33%), 1 patient in TR group (6.66%) and were all managed with endoscopic dilatation successfully. Respiratory complications, mainly post-operative chest infection and pneumonia, included 3 patients in COR group and 2 patients in TR group. Mortality, as a potential complication, was more prevalent in COR group (2 patients, 13.33%) than TR group (1 patient, 6.66%). Death was mainly attributed to sepsis and respiratory failure.

Regarding post-operative scar evaluation, TR group had a highly significant better cosmetic appearance, lower scar score (nearly matching normal surrounding skin) when evaluated both by the patient parents (P-SAS) and the observer surgeon (O-SAS) (p=0.00) (Table 3).

3. Discussion

Thoracotomy is considered standard surgery for repair of esophageal atresia. With advances in pediatric endoscopic surgery and anesthesia, the indications for a thoracoscopic approach are increasing. Several studies were performed to evaluate the effectiveness and safety of TR [10–12] reporting better visualization of posterior mediastinal structure and avoiding skeletal deformities resulting from thoracotomy. Yet, TR is known to be a highly demanding operation requiring high surgical skills due to manipulations within the small chest cavity of a neonate.

EA/TEF occurs in boys more than girls at a rate of 1.26:1 [13]. In the present study, the overall male to female ratio was 1.3:1. Birth weight as a risk factor for survival of neonates with EA has been addressed in numerous studies and low birth weight was identified as risk factor for survival [14]. In the present study, the birth weight ranged from 2400g to 3750g in both groups. It is worth mentioning that a cut-off value of bodyweight for thoracoscopic repair varies from 1500 to 3000 g based on the surgeon's expertise [15].

The age at presentation in the present study ranged from 1 to 12 days in COR group and from 1 to 24 days in TR group. This may be attributed to delayed diagnosis by primary health care providers and pediatricians, shortage of surgical NICU beds in comparison with number of surgical neonates, lack of adequate number of tertiary centers with pediatric surgery capabilities.

EA is frequently associated with other congenital anomalies, most commonly are those present in the VACTERL association [16].

Table 3
Post-operative course and early complications in both groups.

Variable	COR group (n = 15)	TR group (n = 15)	Test	p-Value
Extubation (days)				
Mean + SD (Range)	1.93±1.62 (1-5)	1.53±0.83 (1-3)	MW	0.87
Reintubation				
Number (%)	2 (13.33%)	1 (6.66%)	X2	0.54
First feeding (days)				
Mean + SD (Range)	6.76±1.09 (5-9)	5.93±1.48 (4-8)	T-test	0.20
Hospital stay (days)				
Mean + SD (Range)	11.73±5.68 (9-30)	9.2±2.95 (6-18)	MW	0.07
Complications				
Anastomotic leak	2 (13.33%)	2 (13.33%)		1.00
Stricture	2 (13.33%)	1 (6.66%)		0.54
Wound infection	3 (20%)	0 (0%)	X ²	0.07
Skin dehiscence	3 (20%)	0 (0%)		0.07
Recurrent TEF	0 (0%)	0 (0%)		—
Resp. complications	3 (20%)	2 (13.33%)		0.62
Mortality	2 (13.33%)	1 (6.66%)		0.54
Scar scoring				
P-SAS				
Mean + SD (Range)	3.65±1.07 (2.25-5.5)	1.31±0.40 (1-2.5)	T-test	0.00** (HS)
O-SAS				
Mean + SD (Range)	3.52±1.11 (2.33-5)	1.28±0.32 (1-2)	T-test	0.00** (HS)

SD: Standard Deviation; P-SAS: Patient Scar Assessment Scale; O-SAS: Observer Scar Assessment Scale; COR: Conventional Open Repair; TR: Thoracoscopic Repair; MW: Mann-Whitney test.

** HS: Highly significant (p<0.01).

Cardiovascular anomalies are the most commonly reported associated anomalies with EA/TEF with reported incidence of 11–49% [17]. The present study showed an overall prevalence of cardiac anomalies in 83.33% (25 out of 30 patients), most commonly PDA (43.3%), PFO (40%) and ASD (33.3%). Genitourinary anomalies occurred in 13.33%, gastrointestinal in 6.66%, skeletal and vertebral anomalies didn't occur in any patient (0%) probably due to the small sample size included in the study. Moreover, genetic anomalies occurred in 6.66% in the form of two cases with Down syndrome (trisomy 21).

Several earlier studies compared the operative time in both techniques. One study showed a longer operative time in the COR group [18], whereas many other studies reported that the operative time of TR was longer [6,7,20–23].

In the present study, the results showed a non-significant relatively longer operative time in TR group compared to COR group. This longer operative time in TR group can be attributed to the still developing learning curve with thoracoscopic surgery and mastering the intracorporeal knotting. In the standard technique for repair of EA and tracheoesophageal fistula, the azygos vein is ligated and divided early in the operation. The present study proved a significant ability to preserve the vein in TR unlike COR where it was sacrificed in 33.33%. Sharma et al. [24] proposed that preserving the azygos vein reduces the anastomotic edema by maintaining the venous drainage. The gap length between the upper and lower esophageal pouches is an important predictive factor for successful primary repair. The wider the gap length, the more likely strictures may develop [25], however, this theory was challenged recently [26,27].

The reported rate of conversion in literature ranges from 4% to 44% [3,23]. In the current series, only one patient was converted to open surgery (6.66%) due to injury of the left main bronchus and repair was done and procedure completed in open fashion. Longer time to extubation in COR group was reported by multiple authors [4,6,7,21], however, results of comparative studies regarding time to first oral feeding was variable from superiority of TR [4,6,7] to similarity [28].

there was a general agreement that hospitalization was longer in COR group compared to TR group [4,6,7,19,21].

The most common complications after repair of esophageal atresia and tracheoesophageal fistula relate to the anastomosis. Observed rate of leakage varied from as low as 2.5% to as high as 33.5% [18,22,23]. Meta-analysis studies performed by Borruto et al. [8], Yang et al. [4] and Wu et al. [28] concluded that both COR and TR have similar leakage and stricture rates with comparable outcomes. In the present study, anastomotic stricture occurred more frequently in COR group than in TR group. Similar results were published by different authors [7,18,29]. Both groups in the present study had evidence of respiratory complications in 20% in COR group and 13.33% in TR group.

Koga et al. [7] who revealed high rate of occurrence of repeated pneumonia and atelectasis after open repair and explained it by the compression needed for visualization in COR resulting in mucosal plugging in airway unlike TR where the insufflation by Co₂ lead to uniform parenchymal lung collapse.

In-hospital mortality rate in the present study was 10%, with 2 patients in COR group and 1 in TR group, and this corresponds to the increased respiratory complications in COR. Regarding cosmetic outcome, Lawal et al. [30] found statistically significant advantages in the group who underwent thoracoscopy including less chest asymmetry, less nipple asymmetry, more favorable Manchester scar assessment and more favorable patient satisfaction scores. The present study used POSAS score for scar evaluation showed highly significant better cosmetic appearance, lower scar score when evaluated both by the patient parents and the observer. Slater and Rothenberg [31] concluded that TR is associated with improved cosmesis due to smaller incisions.

4. Conclusion

Thoracoscopic repair of EA/TEF is a safe, effective, minimal invasive technique with comparable outcomes, operative time and complication rate when compared with COR. Conversion rate in TR is minimal (6.66%), justifying its choice as a first line technique for repair of EA/TEF in equipped facilities with experienced surgeons. Moreover, Azygos vein can be significantly spared in TR (100%), unlike COR when ligation may be required for better visualization. Cosmetic outcome is significantly better with TR than with COR, as documented by both parents and surgeons scar assessment scale.

Compliance with ethical standards

Funding

No funding was received for this study.

Conflict of Interest

All authors declare that they have no conflicts of interest.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study.

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