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ORIGINAL RESEARCH

A comparative study of thoracoscopic and open tracheoesophageal fistula and esophageal atresia repair as first single center experience in Turkey

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Abstract

Purpose The focus of esophageal atresia treatment is now on reducing hospitalization and common morbidities and to prevent complications such as anastomotic leak and esophageal stricture formation. The objective of this study was to compare the perioperative and postoperative outcome measures such as operation times, anastomotic leak and stricture formation in children who underwent thoracoscopic and open repair of esophageal atresia/tracheoesophageal fistula as newborns in a single institution.

Methods Children who underwent surgery for esophageal atresia/tracheoesophageal fistula between 2014 and 2020 were enrolled to the study. Charts of patients were reviewed retrospectively for demographic and perioperative data. Postoperative outcome endpoints such as stricture formation rates, anastomotic leak more than 1 week, and of the chest tube were compared.

Results There were 22 children in the study, of which 14 underwent thoracoscopic repair and 8 underwent open repair. Both groups were similar in terms of gestational age, birth weight and associated major anomalies. Operative times were similar (thoracoscopic repair: 169 and open repair: 175 min). Stricture formation was observed in three children in thoracoscopic group (21%) and four children in open group (50%). Anastomotic leak persisted for more than a week in three children in thoracoscopic group (25%) and two in open group (25%). Stricture rates and anastomotic leak for more than one week rates were statistically similar between groups. The azygous veins were preserved in 11 of 14 children in thoracoscopic group while none in the eight children that were operated in open group.

Conclusion Thoracoscopic repair is comparative to open repair regarding operative time and postoperative outcome endpoints such as esophageal stricture formation and anastomotic leak more than 1 week in the study. Thoracoscopic repairs are as feasible as open repairs with the advantage of preserving the azygos vein.

Keywords Esophageal atresia · Newborn · Tracheoesophageal fistula · Thoracoscopy · Thoracotomy

Introduction

Esophageal atresia (EA) is a rare congenital anomaly and may be present one in 5000 newborns [1]. It may be ısolated or with a tracheoesophageal fistula (TEF/EA). In the last century, survival rates of these children have increased to 92% from 0% [2]. In 1941, Haight performed the first successful open repair for a neonate [3]. From that day to 1999; in which the first thoracoscopic repair (TR) was performed; open repair (OR) of TEF/EA remained the gold standard procedure [4]. After the first repair, TR is being performed all over the world and is one of the most challenging procedures for pediatric endoscopic surgeons [1].

Nevertheless better understanding of the newborn physiology during anesthesia, smaller and more convenient endoscopic devices and increased experience of surgeons has led to more TR being performed worldwide [5–7]. Since mortality usually occurs in the presence of major accompanying comorbidities; the focus of EA treatment now is on reducing hospitalization and common morbidities and complications

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such as esophageal strictures (ES), gastroesophageal reflux disease (GERD), musculoskeletal deformities and pulmonary issues [1, 8, 9]. Even though there are reports comparing OR and TR; they remain head to head in terms of many of these aspects [8, 10–12].The primary objective of this analysis was to compare the outcome measures of TR and OR of EA/TEF in newborns in a single institution.

Materials and methods

The study was conducted in accordance with the latest version of the Declaration of Helsinki. Ethical committee permission was granted and all parents were informed about the use of their children's data for scientific purposes.

Children who were operated for any type of EA between 2014 and 2020 and who underwent esophageal atresia repair were included in the study. At the end of 2015, TR was introduced and since then all patients with TEF EA underwent TR. All patients participating in the study prior to this underwent open EA repair. Charts of patients were reviewed retrospectively and demographic and perioperative data were recorded. Children who had long gap atresia and underwent esophageal replacement procedures such as colonic interposition or gastric pull up were excluded.

Preoperative assessment: All children had detailed physical examination by neonatologists, pediatric surgeons and anesthesiologists. Upper pouch contrast study with 0.5 ml water soluble iodine was used to determine the approximate distance between both esophageal ends. Renal ultrasound imaging and echocardiography were also performed concerning associated anomalies in all patients.

Surgery

Preoperative bronchoscopy: Even though the role of preoperative bronchoscopy is controversial in EA surgery, in our center, we started to perform routine preoperative bronchoscopy for the last 4 cases. Bronchoscopy may be beneficial for two reasons; to rule out a proximal fistula and to estimate the gap between two ends of esophagus.

Open repair: The newborn was positioned for a standard right posterolateral thoracotomy and the right arm was placed above the head. Curved skin incision was made from the angle of the scapula and extended to the anterior axillary line. Subcutaneous tissue and muscles of the chest wall were divided. The thoracic cavity was entered through the fourth intercostal space by dividing intercostal muscles. Transpleural or extrapleural approach was opted. The azygos vein was exposed and divided between ties as the lung are gently retracted medially. Posterior mediastinum was exposed and the upper pouch, distal tracheoesophageal fistula and vagus nerve were identified. The distal part of the esophagus was dissected from the level of the fistula. The fistula was closed by 5/0 absorbable suture in interrupted fashion leaving 2 mm esophageal tissue. Tracheal closure was controlled for an air leak after filling the thoracic cavity with saline during positive pressure ventilation. Distal part of the esophagus was dissected carefully for low-tension anastomosis. The upper pouch was identified and dissected until ensuring to bring upper pouch to the distal esophageal part for low-tension anastomosis. The tip of the upper esophageal pouch was excised after a nasogastric tube was gently placed to the upper pouch. The posterior wall was anastomosed using interrupted 5/0 absorbable sutures. The anterior wall of anastomosis was completed after a nasogastric tube was passed through. A chest tube was placed to posterior side of anastomosis and the incision was closed in traditional fashion.

Thoracoscopic repair: The infant was placed near-prone position with the right side slightly elevated. Initially a 4-mm camera port was placed at fifth intercostal space behind the tip of the scapula and 30° telescope was used. Thoracic cavity was insufflated by 1–2 L/min CO_2 with maximum pressure of 4–6 mmHg. Two 3-mm work ports were placed, one at the junction of the seventh intercostal space and the paravertebral space, and the other from the junction of the seventh intercostal space and the anterior axillary line. The azygos vein was exposed and preserved. Tracheoesophageal fistula was dissected and ligated leaving approximately 2 mm esogageal tissue with a metal clip. A nasogastric tube was placed and pushed by the anesthesiologist to identify the upper pouch. Then the proximal esophagus was mobilized until the low-tension anastomosis was possible. A small part of distal esophagus was resected. The tip of the proximal esophagus was opened by scissors. The anastomosis was performed by 5/0 absorbable interrupted sutures above the azygos vein. NG tube was inserted through the distal esophagus into the stomach after first two stitches. A chest tube was placed under direct visualization through the paravertebral port site with the tip of tube being placed beside the anastomosis. The port incisions were closed.

Postoperative period: All patients were managed in the neonatal intensive care unit (NICU) postoperatively until the extubation was performed successfully and stabilized. After eliminating the need for oxygen support, the neonates were transferred to the pediatric surgery department. If the contrast study in the first postoperative week did not show any stricture or leakage, the nasogastric tube was removed and oral feeds were started. After full enteral feeding was established, the feeding of the neonates was organized and the parents received guidance by a multidisciplinary team which included pediatric surgeons, feeding nurses and swallow therapists which is crucial in the postoperative period. Routine controls were first week after discharge to check the feeding and weight gain. If there were no symptoms

in the follow-up, the gap between the outpatient visits was extended, and the parents were informed about the possible difficulty in swallowing and stenosis at the anastomosis site. All children were referred to the pediatric department to be followed up in terms of neurodevelopmental status, respiratory functions and accompanying anomalies.

Definitions

Major associated anomaly: Life threatening anomalies such as cardiac, renal or neural defects that may occasionally be seen occasionally with EA.

Prolonged leak: Anastomotic leak lasting more than a week and evident on contrast study.

Stricture: Stenosis at the level of the anastomosis confirmed by contrast study or evident by symptoms after contrast study and proven by esophagoscopy.

Statistical method

Statistical analysis of data was performed via SPSS for Windows 11.5. Chi-square test was performed in case of comparision of categorical data between two groups. Scaled data comparision was performed via independent *t* test and and Mann Whitney *U* test according to whether they distributed normal or not. *P* values under 0.05 were accepted statistically significant.

Results

There were 30 neonates that underwent surgery for EA between January 2014 and January 2020. Neonated who had isolated EA (IEA) and who underwent esophageal replacement procedures were excluded from the study ($n=8$). Among remaining neonates, 14 underwent TR and eight underwent OR in both groups were statistically similar regarding age, sex and birth weight ($p>0.05$).

Patient characteristics were presented in Table 1.

Preoperative bronchoscopy was performed in four patients to reveal a possible proximal fistula and determine the level of the fistula opening. Proximal fistula was absent in these four patients.

Intraoperative and postoperative data and outcomes were compared. Data on operation time, intraoperative findings, perioperative complications, stricture and postoperative antireflux surgery rates and mortality were also evaluated. Detailed information was shown in Table 2.

Four infants died in the first year after surgery. None of the deaths were related to EA or surgical complications except in one child from the OR group was lost due to apnea caused by severe tracheomalasia after discharge. One of the deaths were in the TR group resulting from cardiac anomalies. (Dextrocardia, secundum ASD). Three of the deaths were in the OR group, 2 because of cardiac anomalies

Table 1 Patient characteristics comparing neonates undergoing thoracoscopic repair (TR) and open repair (OR) for esophageal atresia

	TR ($n=14$)	OR ($n=8$)	*P* value
Age(days)	2.1 (1–3.3)	2.2 (1–3.6)	0.750
Sex (male, %)	64 ($n=9$)	25 ($n=2$)	0.145
Mean gestational age (weeks)	37.4 (35–40)	37.3 (33–40)	0.673
Prematurity, (%)	43 ($n=6$)	50 ($n=4$)	0.752
Median birth weight (grams)	2693 (2300–3560)	24,653 (1950–3625)	0.410
Associated major anomalies (%)	21 ($n=3$)	25 ($n=2$)	0.851
Mean follow-up (months)	18 (12–36)	24 (12–42)	

TR Thoracoscopic repair, *OR* Open repair

Table 2 Intraoperative data and postoperative outcome

Perioperative findings and outcomes	TR	OR	*P* value
Azygos vein preservation	11	0	0.001
Operation time (Mean, minute)	169 (90–360)	175 (75–310)	0.684
Extubation time (postoperative day)	5.15 (1–13)*	5.75 (1–9)	0.725
Prolonged leak (%)	21.4 ($n=3$)	25 ($n=2$)	0.920
Full oral feeding at (postoperative day)	7.9 (3–25)	9.5 (3–26)	0.796
Stricture at postoperative contrast study (%)	21.4 ($n=3$)	50% ($n=4$)	0.297
Strictures requiring more than 3 dilatations	30% ($n=4$)*	40% ($n=2$)*	0.775
Antireflux surgery	7.6% ($n=1$)*	20% ($n=1$)*	0.703

TR Thoracoscopic repair, *OR* Open repair

*Excluded children who died during the postoperative course

(Tetralogy of Fallot, Complete AVSD), and one from apnea (due to tracheomalasia). The infant with tracheomalacia was under control and followed by pediatric pulmonology and pediatric surgery departments but died at home months after the discharge. There were no intraoperative complications in both groups. Prolonged leakage occurred in three and two patients in TR and OR groups, respectively.

Discussion

According to the present study, TR and OR of EA/TEF seem to be equally feasible. Intraoperative findings and postoperative outcomes were statistically similar between two groups.

Isolated EA and long gap EA are still challenging for both surgeons and patients. Different treatment options including thoracoscopy, thoracotomy and even laparotomy have been presented and a major difference between many methods has yet to be identified [13–16]. On the other hand due to technological developments of surgical devices recently, TR has gained popularity after thoracotomy had been accepted as gold standard surgical approach for many years in newborns with EA/TEF [5, 6, 17]. When the first successful EA/TEF repair was carried out by Haight in 1941, survival of the baby was unusual; but nowadays survival rates of babies with EA have increased over 90% [1, 3]. The increase in the rates may be related to increase in surgical practice, but it would be more appropriate to attribute these rates with advanced care being offered in neonatal intensive care units (NICU) and cumulative knowledge in anesthetical approach [6, 7]. Mortality in EA is now determined by different factors such as cardiac and chromosomal anomalies or prematurity rather than EA itself [1]. Since surgery related mortality has become a minor concern; pediatric surgeons have investigated ways to decrease morbidity arising from the operation. Postoperative complications of OR are well described and includes scoliosis, chest wall deformities, esophageal strictures, thoracic nerve injury serious pain and large scars [18, 19]. Another major pitfall of open surgery is intraoperative insufficiency of adequate exposure irrespective whether the operation is performed by extrapleural or transpleural approach. Thus TR was introduced to EA surgery in 1999 to improve at least some of these aspects [5]. It certainly offers better visualization and cosmetic results, but other aspects still remain controversial [19].

One of the most common complications in EA surgery is esophageal strictures which may reduce the quality of life of the patients and cause developmental problems [20]. Wu et al. presented similar stricture rates after TR and OR in a meta-analysis in 2017 [21]. Concordant to this review of the literature, no difference was found regarding the development of stricture formation after TR or OR ($p = 0.176$). The stricture rates of the present study were in the limits of our literature research (30–40%) with 21% for TR and 50% for OR [18]. Even though the rates of TR were lower, the statistical analysis showed no difference ($p = 0.176$). When the dilatations of these strictures were evaluated, it was observed that almost all of them required more than 3 dilatations (85%, $n = 6/7$). But fortunately the children responded well to the dilatation regimens and no other intervention was required. Anastomotic leak is also a common problem and is reported in 12–22% patients [18]. Prolonged leakeage may delay feeding of the children and cause longer hospitalization. Preserving the azygos vein has been reported to be a protective maneuver for leaks [22]. As it was the only statistically significant difference in our study ($p = 0.001$), mostly the preservation of the vein is only possible in thoracoscopic approach. Preserving the azygos vein is also hyothetized in preventing recurrent fistula formation [22].In our series, even though prolonged leakage rates were similar in both groups (21.4 vs 25%, $p = 0.851$) they resolved spontaneously with no need of intervention and recurrent fistula formation did not occur in any of the groups.

The major challenge of the TR is the surgical technique itself, especially the anastomosis. The procedure is still considered as one of the most surgically demanding procedures among pediatric surgical interventions [4, 22]. This level of difficulty and relatively new adoptance of the technique comes along with longer operative times than conventional open approach [8, 21]. In the present study, no significant difference between operative times was observed (169 vs 175 min, $p = 0.684$). The reason for this may be that TR has been introduced after the surgeons had acquired advanced endoscopic surgical skills. In addition, the authors think that changing the position of the baby on the operating table to almost-prone position after the initial cases of TR has ensured a better exposure than lateral decubitis.

TR and OR in the present series were similar in terms of extubation or full oral feeding time. Intraoperative complication or conversion to open surgery were not observed. There was one death in TR group and three in OR. None of the deaths were due to surgery or EA itself. The deaths were attributed to cardiac anomalies in three children and severe tracheomalasia in one. In the present series patient selection only depended on the two surgical teams; with one team having preference for TR and other OR. Due to the low number of cases it was not possible to define specific criteria for neonates who would undergo TR as Yamoto et al., but we strongly believe that such selection may result in improved outcomes [11].

Musculoskeletal deformities after thoracotomy and neurodevelopmental deficiencies are two long term complications of EA repair. Even though Laberge and Blair claimed that an appropriate thoracotomy has no such risk more than thoracoscopy, scoliosis after thoracotomy reported

repeatedly contrarily to TR [19, 23–25]. Neurodevelopmental delay was related to anesthesia exposure time and possible decrease of brain oxygenation during anesthesia in the neonatal period [26]. Bishay et al. performed the only randomized controlled study on patients with EA that underwent open and thoracoscopic surgery and they found no difference in terms of hypercapnia and acidosis even though they stated children underwent thoracoscopic congenital diaphragm hernia repair experienced longer durations of extreme hypercapnia and acidosis levels [27].

Retrospective nature with limited number of patients is a limitation of the study. Nevertheless considering rarity of the disease and numbers in other studies, these amount of patients seems average [11, 12, 28]. Also aforementioned benefits of the almost-prone position were not documented and only depends on clinical observations. The study only includes short gap EA to perform a standardized comparison since primary anastomosis may rarely be possible in long gap cases. Also, not having data on long term complications should be mentioned as a limitation of the present study even though another well designed study should address on these aspects.

Conclusion

Although TR of EA requires advanced endoscopic surgical skills, the outcomes are similar to OR regarding operation time and postoperative outcomes such as esophageal stricture formation and prolonged leakage in this study. Randomized controlled studies with larger patient groups addressing these aspects may offer objective perspectives. Prospective studies with longer follow-up periods are required to fill the gap in data.

Declarations

Conflict of interest None.

Financial disclosure There were no financial or non-financial interests that are directly or indirectly related to the work.

Ethical approval Ethical approval obtained.

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