



Comparison of Two Transpleural and Extrapleural Surgery Methods in Neonatal Esophageal Atresia

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Abstract

Background: Esophageal Atresia and fistula is a congenital anomaly and its treatment is surgically via thoracotomy or thoracoscopy. Its subject of debate that which kind of surgery and esophageal exposure is better, Extrapleural without opening of pleura or transpleural with opening of pleura. It seems that extrapleural route had better prognosis if anastomosis leak occurs and most surgeons prefer this method. In this study we compare the two methods and its results.

Material and Methods: In this retrospective study, 72 neonates in Imam Reza and Mohammad Kermanshahi hospital between March 2007 and March 2014 had been operated due to esophageal atresia (36 male and 36 female neonates) 39 patients with extrapleural and 33 patients with intrapleural method.

Results: Seven anastomosis leaks happened after surgery, from which 6 cases were in intrapleural group. Totally 20 cases died, from which 8 cases were related to extrapleural method and 12 cases from transpleural group. There were 4 cases of T.E. fistula recurrences after surgery from which 3 cases were belongs to intrapleural group (one case died) and only one case from extra pleural group.

Conclusion: Esophageal anastomosis leak after surgery for esophageal atresia in extra pleural method is lesser than intrapleural method and if it occurred, prognosis is better in extrapleural method, probably, due to intact pleura and absence of any relation between leakage site and mediastinum that prohibits infection to spreading within lung and another organ. In addition, better drainage with chest tube in extrapleural method is important in faster repair and improvement.

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Keywords: Esophageal atresia; Transpleural repair; Extrapleural repair; Leak of anastomosis

Introduction

Esophageal atresia is a congenital defect. There are several types. In most cases, the upper esophagus ends and does not connect with the lower esophagus and stomach. The top end of the lower esophagus connects to the windpipe; this connection is called a Tracheoesophageal Fistula (TEF).

Esophageal atresia occurs in about 1 out of 4,000 births. Symptoms are Drooling and Cyanosis, Coughing, Gagging, and choking with attempted feedings. Before birth, an ultrasound performed on the pregnant mother may show too much amniotic fluid, which can be a sign of esophageal atresia or other blockage of the digestive tract. The disorder is usually detected shortly after birth when feeding is attempted and the infant coughs, chokes, and turns blue. As soon as the diagnosis is suspected, an attempt to pass a small feeding tube through the mouth or nose into the stomach should be made. The feeding tube will not be able to pass all the way to the stomach in a baby with esophageal atresia. An X-ray of the esophagus shows an air-filled pouch and air in the stomach and intestine (Figure 1). If a feeding tube has been inserted, it will appear coiled up in the upper esophagus. Esophageal atresia is considered a surgical emergency. Surgery to repair the esophagus should be done quickly after the baby is stabilized so that the lungs are not damaged and the baby can be fed. Surgery is almost always done soon after birth [1]. Some children do experience problems following TEF surgery; they can develop dysphagia and thoracic problems. Children with TEF can also be born with other abnormalities; most commonly those described in VACTERL association a group of anomalies which often occur together, including heart, kidney and limb deformities. Classification: Fistulae between the trachea and esophagus in the newborn can be of diverse morphology and anatomical location; and based on this, it divides into 5 subtypes [2]. Treatment: Tracheoesophageal fistula and esophageal atresia can usually be repaired at the same time. The surgeon will make a cut on the right side of the chest between the ribs. After thoracotomy, esophagus can be approached

via extrapleural or transpleural route, another less invasive method of esophageal repair is by thoracoscopy, without opening the thorax. In all methods, however, the fistula, which is the abnormal connection between the esophagus and windpipe, is closed off. Then the upper and lower portions of the esophagus are sewn together. If the 2 parts of the esophagus are too far apart, then, only the fistula will be repaired during the first surgery. A gastrostomy tube (a tube that goes through the skin into the stomach) may be placed to give your child nutrition [3]. As with any surgery, complications may arise after the procedure. These include: Esophageal Leak, this usually heals over several weeks if minor. If more significant, the esophagus may require re-repair or diversion of the esophagus into the neck. Gastroesophageal reflux treated with antacids but may require a surgical anti-reflux procedure in the future. Tracheomalacia: Barking type cough. Cyanosis immediately after feedings. Onset is usually 2 to 3 months post-op, if severe may require a procedure to lift the trachea away from the esophagus. Poor esophageal motility is common. It generally improves with time but may require medical treatment or dietary modification.

Esophageal Strictures may develop in many patients and can be managed with outpatient dilations if uncomplicated. More severe strictures may require resection of the stricture. Recurrent TEF fistulas may require additional procedures to remove the recurrent fistula. Dysphagia, difficulty swallowing that generally improves over time and may require intensive swallow therapy [4]. Which method of surgery is better, its subject of debate. Most pediatric surgeons would prefer to correct esophageal atresia using an open extrapleural approach, the technique of an extrapleural approach has many potential benefits, the real reason is to make sure that if esophageal anastomosis leak occurs, and it would actually be contained to the extrapleural space and less dangerous. Another reason to choose extrapleural route is that, if we can do the initial repair extrapleurally and complication happens, future transpleural procedures can become technically easier [5]. In this study we compare results and complications of two methods of open esophageal repair surgery (extrapleural without opening the pleura or transpleural with opening the pleura).

Materials and Method

This study was performed between March 2007 and March 2014 in the Kermanshah University of Medical Sciences Pediatric Surgery Center (Mohammad Kermanshahi and Emamreza Hospitals). As a randomized, single-blind clinical trial 72 patients with esophageal atresia type-c were included in the study (39 cases in extrapleural and 33 cases in transpleural group). Allocation of the all cases were random, depending on intra operative condition and how the surgeon could be able to keep the pleural layer intact or opening the pleural layer inadvertently, and namely all subject were assigned to a treatment group (extrapleural) or control group (transpleural). All patients with other types of malformation (types A-B-D-E-H.) were excluded from the study. All patients under 2 kilogram weight and patients with long gap atresia excluded also. The presence of other possible accompanying anomalies was determined by echocardiography and abdominal sonography. All cases underwent surgery in the first few days of life. All surgeries were performed by one pediatric surgeon. The parents of all children in the study signed an informed consent form that contained the necessary information. The study was approved by the Ethics Committee of the University. In the control group (extrapleural repair), the neonates underwent



Table 1: Compare of tracheoesophageal fistula recurrence in 2 groups.

CI	RR	P-value	Percent	Number	Group
Lower=0.387	54/3	0.22	9/1	3	Transpleural
Upper=32/48			2/6	1	Extrapleural

a right posterolateral thoracotomy and esophagus exposed *via* dissection of visceral pleura of the lung and pushing it a way without opening pleura and exposing lung. Distal esophagus exposed and tracheoesophageal fistula cuts and repaired, then finding proximal esophagus and freeing it, anastomosis of distal and proximal segments of esophagus and finally insertion of a chest tube in retropleural space as a drain and finally closing the thorax. If pleura ruptured inadvertently, then repair of esophagus and its fistula to trachea has been done transpleurally and chest tube inserted in pleural space and surgery terminated. Patients with intact pleura remained in study group and those with ruptured pleura and entrance into pleural space put in control group. All cases in both groups were kept under close observation in the post-operative period in the hospital, as well as during intermittent outpatient visits in the clinic after discharge. The participants in the study were followed up for several months, (the follow-up after completion of the treatments). Then the final results, duration of operation time, recovery period, mortality and complications were compared between the two groups. The most important complications, including leak of anastomosis, esophageal stricture, recurrence of tracheoesophageal fistula, pneumothorax, and wound infection, compared together.

All data that was obtained collected using a checklist designed to conduct the study. Finally, the data analysis was done using SPSS20

We assessed all criteria and variables like: age, weight, operation time, surgical method, recovery time, and complications like: anastomosis leakage, stricture, fistula recurrence, pneumothorax, wound infection and mortality rate.

We used descriptive statistics to draw charts and graphs, and used independent t-test or Mann-Whitney test for compare of the quantitative variables and chi-square and fisher test and logistic regression for compare of qualitative variable. SPSS20 software is used and $P < 0.05$ will be a significance level.

Results

Seventy two cases of esophageal atresia (type-c) underwent surgery between March 2007 and March 2014 in the Kermanshah University of Medical Sciences Pediatric Surgery Center (Mohammad Kermanshahi and Emamreza Hospitals) 36 male and 36 female

Table 2: Compare of anastomosis leakage in two groups.

CICCI	RR	P value	Percent	Number	Group
Lower=0.089	RR=4.09	P=0.026	18/2	6	Transpleural
Upper=55.94			2/6	1	Extraleural

neonates; 39 patients with extra pleural and 33 patients with intrapleural method of the patients surveyed 20 patients died (27.7 percent mortality rate).

From whom 8 cases were from study group (extra pleural method) and 12 cases were from control group (intrapleural method). Most patients who had died were low birth weight or long gap esophageal atresia that made surgery problematic technically, neonates weighing between 2 kg and 5.3 kg, respectively. And all neonates underwent surgery in the first few days of life by one pediatric surgeon with 20 years experience. Persistent Leak of anastomosis was observed in 7 patients, 6 cases from transpleural and 1 case from extrapleural group. Four patients in transpleural group died due to persistent leakage of esophageal anastomosis and sepsis, but one case of anastomosis leakage in extrapleural group recovered and healing happened rapidly without sepsis. Tracheoesophageal fistula recurrences after surgery occurred in 4 cases, 3 from transpleural and 1 from extrapleural group (Table 1), all of them underwent revision surgery and fistula repaired, one of them finally expired due to surgical complication and other 3 improved and survived. We had only one case of wound infection in extrapleural group who finally improved. We had not any case of severe stricture in both groups also.

Mean duration time of operation in the extrapleural method was 158/33 minute and in transpleural group was 160/9 minute. Difference was not statistically significant.

Note: The operation time considered from the start of anesthesia to the patient waking up and the average calculated time for each patient's surgery lasted very shorter (about 1 hour).

Reviews of anastomosis leakage in the two groups showed 6 cases of anastomosis leakage in transpleural group and 1 case (2/6) percent in (18/2) percent extrapleural group (Table 2). This difference was statistically significant and the incidence of anastomosis leakage in extrapleural method is less than transpleural surgery P=0.026.

Discussion

A review of the experience with esophageal atresia and tracheoesophageal fistula over a 25-year period appears to lead to the advisability of the Extrapleural interruption of tracheoesophageal fistula and end-to-end esophago-esophagostomy in patients who have the common type of upper esophageal atresia with distal tracheoesophageal fistula [6].

One of disadvantage of extrapleural repair of esophageal atresia is need to insertion of second chest tube if pneumothorax or pleural effusion happens. In a review of 110 cases of Esophageal Atresia (EA) and Tracheoesophageal Fistula (TEF), 15 patients (13.6%) developed anastomotic leakage. All patients had extrapleural surgical exposure and Prophylactic Extrapleural Chest Drainage (PEPCD). Despite this precaution, all patients with anastomotic leaks developed a pneumothorax with or without a pleural effusion. This required insertion of an additional drain in 12 (80%) cases. PEPCD following repair of EA and TEF does not appear to prevent pneumothorax or pleural effusion when anastomotic leakage occurs [7]. Another less invasive approach to repair of esophageal atresia is *via* thoracoscopy

Table 3: Compare of mortality rate in 2 groups.

CI	RR	P value	Percent	Number	Group
Lower=0.59	0.8	135/0	36/4	12	Transpleural
Upper=1.08			20/5	8	Extraleural

without opening the thorax. A cohort of international pediatric surgeons from centers that perform advanced laparoscopic and thoracoscopic operations in infants and children retrospectively reviewed their data on primary thoracoscopic repair in 104 newborns with EA/TEF. Newborns with EA without a distal TEF or those with an isolated TEF without EA were excluded. In these 104 patients, the mean operative time was 129.9 min (\pm 55.5), and the mean days of total hospitalization were 18.1 (\pm 18.6). Twelve (11.5%) infants developed an early leak or stricture at the anastomosis and 33 (31.7%) required esophageal dilatation at least once. Five operations (4.8%) were converted to an open thoracotomy and one was staged due to a long gap between the 2 esophageal segments. Twenty-five newborns (24.0%) later required a laparoscopic fundoplication. A recurrent fistula between the esophagus and trachea developed in 2 infants (1.9%). Three patients died, one related to the EA/TEF on the 20th postoperative day. They concluded that thoracoscopic repair of EA/TEF can be safely performed by experienced endoscopic surgeons. The results presented are comparable to previous reports of babies undergoing repair through a thoracotomy. Based on the associated musculoskeletal problems following thoracotomy, there will likely be long-term benefits for babies with this anomaly undergoing the thoracoscopic repair [8].

In another study in United Kingdom on 107 neonates with esophageal atresia, they evaluated the necessity of insertion of an Intercostal Drainage (ICD) after repair of esophageal atresia in two methods of repair. Patients were analyzed in two groups-group 1 (n=73) had an Extrapleural (EP) repair and group 2 (n=34) had a purposeful Transpleural (TP) approach (surgeon preference). Eleven patients (10%) had an operative ICD, of which six patients were in group 1 and five in group 2. These 11 patients had an uncomplicated postoperative course and all operative ICD were removed within 48 h of surgery. Of the 96 patients that did not have an operative ICD, only 2 (2%) required postoperative intervention. One patient, in group 2, had a postoperative ICD inserted for a simple pneumothorax at 12 h and removed at 48 h. The other patient, in group 1, had a clinically detected pneumothorax, they find no differences between two group [9]. In another study in Iran on thirty cases of esophageal atresia, neonates divided in 2 groups, 15 of whom underwent surgery by extrapleural approach and 15 by transpleural approach. Five patients (16.6%) developed leakage of anastomosis, 3 of whom were in the transpleural group and 2 were in the extrapleural group. There were no significant statistic differences in the rate of anastomosis leakage between the surgical techniques. Three patients died after surgery (10%), two of whom were in the transpleural group and one was in the extrapleural group. There were no significant differences in mortality rates between the surgical techniques. Duration of operation was significantly shorter in the transpleural group (Table 3). There were no significant statistic differences between the two groups in duration of hospitalization. In other studies, rate of leakage of anastomosis was 10% to 20% and in a textbook of pediatric surgery, it was 14% to 16% [10].

Conclusion

Leak of anastomosis in esophageal atresia repair is less in

extrapleural repair and if happens the recovery is faster with better result than transpleural method of repair. It may be due to presence of intact pleural layer that acts as a barrier and prohibits of leak diffusion and infection and better drainage of leak by chest tube in this method. It seems further studies with more cases is necessary to resolve the issue.

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