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The learning curve for thoracoscopic repair of esophageal atresia with distal tracheoesophageal fistula: A cumulative sum analysis

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ABSTRACT

Background: Thoracoscopic repair of esophageal atresia with tracheoesophageal fistula (EA/TEF) remains technically challenging due to the rarity of these procedures. The aim of this study is to report our experience with thoracoscopic repair of type C EA/TEF and to evaluate the learning curve based upon the surgeon's skill level.
Methods: We retrospectively reviewed data of thoracoscopic EA/TEF repair performed in our center between October 2008 and May 2019. The learning curve was evaluated using the cumulative sum (CUSUM) method based on operative time.
Results: Of the 50 consecutive cases evaluated, the mean birth weight was 2634 ± 608 g and the median age at operation was 3 days (range, 1–29 days). The mean operation time was 144 ± 65 min. Anastomosis leakage occurred in 3 cases (6%) and strictures requiring balloon dilatations occurred in 16 cases (32%). The CUSUM analysis evaluated a learning curve of approximately 10 cases of thoracoscopic type C EA/TEF repair. A lower gestational age was associated with longer operation time.
Conclusions: Thoracoscopic repair of type C EA/TEF is a feasible and safe procedure. The number of procedures required to achieve a stable learning curve was 10. The learning phase may be shortened by adequate set-up under the supervision of an expert endoscopic surgeon.
Type of study: Retrospective Comparative Treatment Study.
Level of evidence: III

The first thoracoscopic repair of pure esophageal atresia was performed in 1999 and the thoracoscopic repair of esophageal atresia with tracheoesophageal fistula (EA/TEF) was first reported 1 year later [1,2]. Since then, many pediatric surgeons started adopting thoracoscopic approach and numerous reports described the outcomes of thoracoscopic repair of EA/TEF [3]. Thoracoscopic repair of EA/TEF provides several advantages, including a magnified operative view, more favorable cosmetic results, and minimal musculoskeletal morbidity compared with thoracotomy [4]. Unfortunately, the procedure remains technically challenging for most pediatric surgeons due to the rarity of these procedures and the particularly small working space in the thorax. Further, previous reports investigating the learning curve and postoperative outcomes of the procedure focused mostly on a single, well-experienced surgeon. In this regard, comparison of learning curves between experienced surgeon and relatively less experienced surgeon facilitated acquisition of surgical skills for safety and efficacy. The aim of this study is to report the outcomes of our experience and to evaluate the learning curve for thoracoscopic repair of type C esophageal atresia with distal tracheoesophageal fistula (EA/dTEF) based upon the surgeon's skill level.

1. Materials and methods

1.1. Patients and data

At our center, the first case of thoracoscopic repair of EA/dTEF was performed in October 2008. Since then, thoracoscopic repair has been considered as the first surgical method of choice for all infants with EA/dTEF. Between October 2008 and May 2019, 66 infants were diagnosed with EA/TEF and underwent surgical correction at the Samsung Medical Center (Seoul, South Korea). The infants were EA/dTEF (type C) in 62 cases, pure esophageal atresia (type A) in two cases, and tracheoesophageal fistula without esophageal atresia (type H) in two cases.

Thoracoscopic EA/dTEF repair were consecutively planned and carried out by two surgeons in all 62 infants with EA/dTEF during this period at our center. Two infants died on postoperative days 18 and 22 due to cardiac anomalies and were excluded from the analysis. Ten infants who underwent thoracoscopic repair and concurrent combined surgery were also excluded from the analysis because the total

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operation time did not specify the time for only thoracoscopic repair in our medical records. Finally, data from 50 infants were included in the analysis. The annual number of cases performed by two surgeons is shown in Fig. 1.

This study was approved by the Institutional Review Board of Samsung Medical Center (approval number: 2019–10-001) and the need for informed consent was waived.

1.2. Surgical technique

Our group previously described the technique used for thoracoscopic repair of EA/dTEF and postoperative management [5]. The infant is laid in a left semi-prone position under general anesthesia. After insertion of a 5 mm scope trocar just below the tip of the right scapula, the thoracic cavity is inflated with CO_2 gas at a flow rate of 3 mL/min, up to a maximum pressure of 5 mmHg. A 3 mm trocar is inserted at the right mid-axillary line along the scapula margin and another 3 mm trocar is inserted opposite the 5 mm trocar. The azygos vein is then divided using electrocautery. After the proximal esophagus is dissected from the adjacent tissue, the distal esophagus is identified and dissected from the trachea until the fistula is visible. The tracheoesophageal fistula is ligated by suture ligation with 3–0 braided polyglycolic absorbable sutures (Safil®; B. Braun, Melsungen, Germany). Further, anastomosis is done by interrupted suture using 5–0 glyconate monofilament interrupted sutures (Monosyn®; B. Braun, Melsungen, Germany) after insertion of a transanastomotic 8 Fr feeding tube. Finally, a 10 Fr chest tube is routinely inserted with the tip placed around the anastomosis.

Postoperative ventilator weaning is attempted as soon as possible the next day after the operation. Oral intake is restricted until contrast esophagography is performed on the seventh postoperative day. In case of no anastomotic leakage, oral intake is initiated and the chest tube is removed on the following day.

1.3. Statistical analysis

Statistical analysis was performed using IBM SPSS Statistics (version 25.0 for Windows, SPSS Inc., Chicago, IL, USA). A p-value <0.05 was considered statistically significant, and confidence intervals were set at the 95% level. Continuous variables are presented as mean ± standard deviations and categorical variables are presented as the number of cases and percentage (%). Mann–Whitney *U* test was used for continuous variables and the chi-square test or Fisher's exact test was used for categorical variables.

1.4. Cumulative sum method and learning curve analysis

The cumulative sum (CUSUM) method was originally developed to monitor the performance in industrial processes. The CUSUM plot is basically a graphical representation of the outcome trend in a series of consecutive procedures performed over time [6]. Steiner et al. described the CUSUM analytical method to monitor surgical performance [7]. The CUSUM technique is a more sensitive method of surgical performance analysis than the standard statistical techniques. Recently, this method has been used for learning curve analysis. A binary outcome is required for CUSUM analyses, and in this study, the binary outcome was surgical failure and success. According to previous studies in the field of minimally invasive surgery, surgical failure was traditionally defined as conversion to open surgery [8]. However, in this study, surgical failure was defined based on the operation time because there was only a single case of open conversion. Failed surgery was defined by longer procedural time compared with the mean operation time. The one case of open conversion was also classified as surgical failure. Operation time was defined as the time from skin incision to skin suture completion. The mean operation time among the whole cases was set as the cut-off value. In the CUSUM plot, the procedures were ordered chronologically on the horizontal axis and data for each procedure were plotted sequentially on the chart from left to right. The graph ascended for every procedure when the surgery time was above the mean operation time (surgical failure) and descended for every procedure when the surgery time was below the mean operation time (surgical success). As a result, the CUSUM was displayed graphically and showed the cumulative total representing a combination of increments with each surgical failure and decrements with each surgical success. The successful learning curve was seen as a continual decline in the curve from its peak [9].

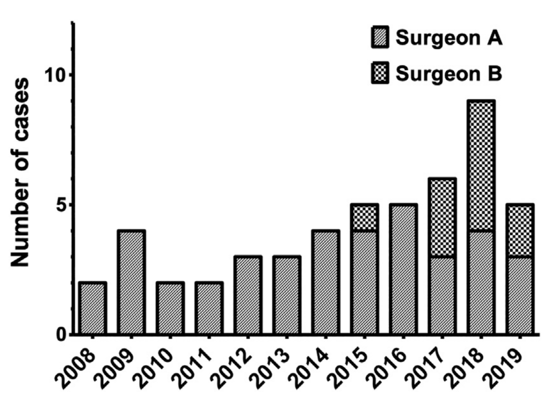

Fig. 1. Annual number of cases.

2. Results

Demographics and surgical outcome including complication of the 50 infants are shown in Table 1. The mean birth weight was 2634 ± 608 g. The median age at operation was 3 days (range, 1–29 days). The mean operation time was 144 ± 65 min. The median postoperative use of mechanical ventilation was 2 days (range, 0–25 days) and the first postoperative day of feeding was 7 days (range, 6–31 days), and 19 days were required to reach full oral feeding (range, 9–305 days). A contrast study was conducted on postoperative day 7. In three leakage cases (3/50, 6%), small amount of localized extra-luminal contrast leakage was found at the esophageal anastomosis site, which then drained into the chest tube. All cases were managed successfully with delayed feeding and spontaneous sealing-up of the leakage sites were confirmed with follow-up contrast studies conducted in one-week intervals.

Table 1
Patient demographics and surgical outcome.

Variables	N = 50
Male:female	35:15
Gestational age (weeks, mean)	37.7
Birth weight, g	2634 ± 608
Age at operation, days	3 (1–29)
Weight at operation, g	2556 ± 583
Operation time, minute	144 ± 65
Associated anomaly	28 (56)
Cardiac anomaly	22 (44)
VACTERL	6 (12)
Postop. MV, days	2 (0–25)
1st oral feeding, days	7 (6–31)
Full oral feeding, days	12 (8–123)
Postop. day of discharge, days	19 (9–305)
Duration of follow up, months	18.6 (1.2–101.3)
Open conversion	1 (2)
Postoperative complications	
Anastomotic leakage	3 (6)
Anastomotic stricture requiring dilation	16 (32)
Number of dilatation	2 (1–8)
GER	19 (38)
GER requiring fundoplication	9 (18)

MV, mechanical ventilation; GER, gastroesophageal reflux.
Data are presented as mean ± standard deviation, median (range), or number (%).

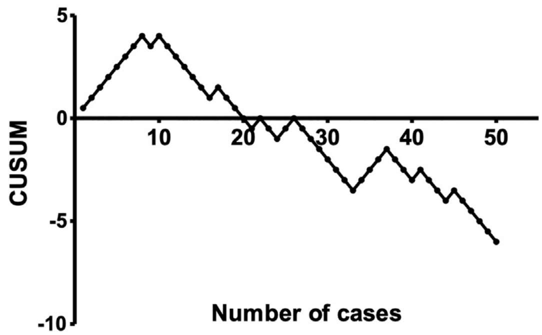

Fig. 2. Cumulative sum plot based on operation time in our center.

Sixteen patients (32%) with clinically significant esophageal strictures showed radiological evidence of anastomotic strictures. All of these 16 patients required one or more sessions of balloon dilatation and the actual median number of balloon dilatations performed was 2 (range, 1–8). No cases of fistula recurrence occurred.

We defined surgical failure by longer procedural time required for completion compared with the mean operation time. The mean operation time was 144 min and 20 cases exceed the mean time to complete. The learning curves were constructed via CUSUM analysis. The CUSUM plot in Fig. 2 reveals that the typical learning curve was accomplished after approximately 10 thoracoscopic procedures. Fig. 3 shows the CUSUM plots of two surgeons in our center individually, labeled as A and B. The surgeons performed 39 and 11 procedures, respectively. The CUSUM plot of surgeon A was similar to the learning curve depicted in Fig. 2. Surgeon B's upward CUSUM plot showed an upward trend initially during the first six procedures followed by a subsequent decline.

Table 2 shows the variables between surgical success and failure groups based on operation time. The gestational age was significantly lower in the surgical failure group ($p = 0.017$).

3. Discussion

Thoracoscopic repair of EA/dTEF is still a technical challenge demanding a high level of surgical skill. In our center, we performed the first case of thoracoscopic EA/dTEF repair in October 2008. Since then, thoracoscopic repair has been considered as the first surgical method of choice for all infants with EA/dTEF in our center. Thus, 62 consecutive thoracoscopic EA/dTEF repairs were performed by two surgeons by May 2019. We previously reported our early experience of thoracoscopic EA/dTEF repair of 22 cases in 2014. In that study, comparing the outcomes during the initial period and the more recent period showed that increased experience shortened the operation time and decreased esophageal stricture [5]. The evaluation of learning curve in that study entailed splitting of patient cohort into sub-groups by study period. This approach is frequently used in numerous studies, however, it has poor sensitivity for the determination of the learning curve transition point [10]. Likewise, other statistical methods for evaluation of surgical learning curve such as various regression models have a limited ability to reveal the characteristic features of the learning curve such as peaks and plateaus. CUSUM plot is a valuable method for the detection of learning curve patterns that otherwise may not be as apparent with simple plots. The application of CUSUM analysis has been increasingly reported in recent studies; however, it has been described in only a small number of publications within the field of pediatric surgery [11].

In order to utilize the CUSUM method for the assessment of surgical performance, a binary outcome measure is required. Most previous studies conventionally defined surgical failure as a conversion to open procedure in field of minimally invasive surgery. However, only one case of open conversion was observed during our study period. Operative time has traditionally been assumed to be the parameter for surgical proficiency and evaluation of the learning curve. Reduced operative time is expected with the increase in experience, although it has a limited role in clinical evaluation. Therefore, surgical failure was defined by longer time for completion compared with the mean operation time in the present study. An upward trend in CUSUM curve was observed for the first 10 procedures, representing the learning phase for surgeon. Thereafter, the curve plateaued and declined continually. This typical learning curve pattern indicates performance at the agreed standard after 10 thoracoscopic procedures. This finding can be compared with that of few other studies discussing the learning curve for thoracoscopic repair of EA/dTEF. Hiradfar et al. reported that the open conversion rate decreased from 58.3% to 35.7% after the first 10 cases [12]. van der Zee et al. compared earlier 41 cases with later 31 cases of thoracoscopic EA repair. Although, the subsequent case series were performed by less experienced surgeons, the mean operation time remained unchanged [13]. Okuyama et al. reported that the operative time decreased significantly as the number of consecutive cases increased [14]. However, the evaluation of surgical learning curve based on these results may be considered inappropriate because the analysis was based on a small study population performed by a single surgeon or comparison of arbitrarily divided sub-groups. The present study is, therefore, the first analysis of a learning curve in a large series of thoracoscopic EA/dTEF repair using the CUSUM technique.

Fig. 3 shows CUSUM plots of two surgeons individually and the CUSUM plot in Fig. 2 represents a learning curve at the institutional level. Surgeon A is an experienced pediatric surgeon with several years of surgical experience in neonatal laparoscopic and thoracoscopic surgery. Fig. 2 and Fig. 3A are similar because most cases involving early study (about 80%) were performed by surgeon A. Data involving surgeon B mainly from 30th procedure onwards may have accounted for the sudden increments observed in Fig. 2. Fig. 3A showed a stable

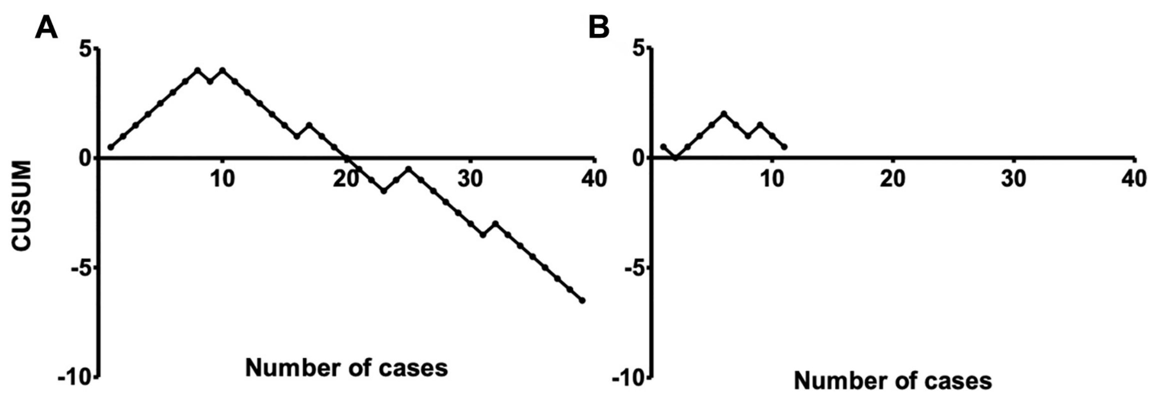

Fig. 3. Cumulative sum plot based on operation time by (A) surgeon A, and (B) surgeon B.

Table 2
Variables in surgical failure and success groups.

Variables, n (%)	Univariable analysis		
	Success (N = 30)	Failure (N = 20)	p-Value
Male	19 (63.3)	16 (80.0)	0.208
Gestational age, weeks	38.3 (2.05)	36.8 (2.14)	0.017
Preterm infant	8 (26.7)	8 (40.0)	0.322
Age at operation, days	3.6	4.3	0.570
Birth weight, g, mean ± SD	2584 ± 504	2710 ± 744	0.478
Weight at operation, g, mean ± SD	2471 ± 498	2684 ± 684	0.208
LBW infant	14 (46.7)	10 (50.0)	0.817
Number of patients who had weight <2.5 kg at operation	18 (60.0)	10 (50.0)	0.485
Associated anomaly	16 (53.3)	12 (60.0)	0.642
Major cardiac anomaly	4 (13.3)	3 (15.0)	1.000
VACTERL	3 (10.0)	3 (15.0)	0.672

SD, standard deviation; LBW, low birth weight.

trend of decline after 10 cases. Surgeon A has performed two to five cases per year steadily and it may affect his learning process as one of external factors for learning curve (Fig. 1).

Previous study showed that supervision by a technically qualified surgeon affects the proficiency of laparoscopic surgery performed by novice surgeons [15]. Similarly, in our analysis, the CUSUM plot for surgeon B appears to decline after six procedures despite a limited number of cases experienced (Fig. 3B). Surgeon B is a pediatric surgeon with limited expertise in thoracoscopic procedures; however, he had completed 2 years of training in minimally invasive surgery under the supervision of surgeon A prior to his first thoracoscopic case. Although it is hard to quantify the effect that supervision by a senior surgeon has on the less experienced surgeon, most pediatric surgeons would strongly agree that practical tips on technical skill and overall mentorship by an experienced surgeon will aid in reducing the learning curve of complex minimally invasive surgical procedures. This mentorship by surgeon A, along with the protocolized surgical set-up and cumulative experience of the surgical team (including anesthesiology and nursing staff), may attribute to the shortened learning phase of surgeon B seen in this study.

Overall, only a single case required conversion to open thoracotomy in our early experience because of inability to sustain adequate oxygen saturation after CO_2 insufflation and subsequent collapse of the right lung. The rate of anastomotic leakage was 6% and the leak resolved with conservative management in all cases. The anastomotic stricture rate in our study was 32%, which is comparable to previous reports [16]. Esophageal stricture was treated with intermittent esophageal balloon dilatations, based on relief of symptoms and radiological findings. Three patients required more than six sessions of balloon dilatation, and another 13 patients were managed via only one to two sessions. The mean operation time was 144 ± 65 min in our study (Table 1). Previous studies reported that the average (median) operative time of thoracoscopic repair for EA/dTEF ranged from 100 to 230 min [17].

According to previous studies, anatomical factors such as small body weight, tiny distal esophagus, and long gaps accounted for the longer operation time [14]. As shown in Table 2, gestational age was significantly lower in the surgical failure group in our study. The lower gestational age was associated with longer operation time. Previous studies reported ventilation difficulty during anesthetic maintenance of EA/dTEF repair, especially in infants with cardiac anomaly, low birth weight, and low gestational age [18]. Lower gestational age infants with a more immature system may lead to poor oxygen saturation after CO_2 insufflation and subsequent collapse of the right lung. During surgery, therefore, repetitive and temporary cessation of CO_2 insufflation to recover oxygen saturation prolonged the operation time.

There were several limitations to this study. First, it was a nonrandomized retrospective cohort study, and therefore the patient information in our data may be incomplete. Second, data in this study involved more than a single surgeon, leading to operator-dependent bias. Lastly, we performed the CUSUM analysis based on operative time as the sole surgical outcome. Further studies will be required to identify the learning curve based on other clinical parameters such as surgical outcomes and postoperative complications including anastomotic stricture and leakage.

In conclusion, thoracoscopic repair of EA/dTEF is a feasible and safe procedure. This study demonstrated that at least 10 thoracoscopic EA/dTEF repair procedures were required to achieve a stable learning curve. However, the learning phase may be shortened when a less experienced surgeon performs the procedure under the supervision of an experienced endoscopic surgeon.

Disclosures

Wontae Kim, Joonhyuk Son, Sanghoon Lee, and Jeong-Meen Seo have no conflicts of interest or financial ties to disclose.

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