Fiberoptic bronchoesophagoscopy-assisted evaluation and prognostic factor analysis in children with congenital esophageal atresia and tracheoesophageal fistula

Chia-Sui Chou[a,b,c], Wen-Jue Soong[c,d], Yu-Sheng Lee[a,c,e], Pei-Chen Tsao[a,c,e], Wei-Yu Chen [a,c,e], Szu-Jung Chen[a], Chin-Su Liu[c,e,f], Mei-Jy Jeng[a,c,e,*]

[a]Institute of Emergency and Critical Care Medicine, National Yang-Ming University, Taipei, Taiwan, ROC; [b]Department of Pediatrics, Taipei Veterans General Hospital, Taoyuan branch, Taoyuan, Taiwan, ROC; [c]Department of Pediatrics, Children's Medical Center, Taipei Veterans General Hospital, Taipei, Taiwan, ROC; [d]Department of Pediatric Pulmonology, China Medical University Children's Hospital, Taichung, Taiwan, ROC; [e]Faculty of Medicine, School of Medicine, National Yang-Ming University, Taipei, Taiwan, ROC; [f]Section of Pediatric Surgery, Department of Surgery, Taipei Veterans General Hospital, Taipei, Taiwan, ROC

Abstract

Background: Esophageal atresia (EA) and tracheoesophageal fistula (TEF) are serious congenital anomalies with high morbidity and mortality. Diagnostic and therapeutic fiberoptic endoscopy has been used in children to evaluate and manage tracheaesophageal anomalies. This study aimed to evaluate the prognostic factors and the role of fiberoptic bronchoesophagoscopy (FB) in managing children with EA and TEF.

Methods: From 2000 to 2017, hospitalized children with suspected EA and TEF were enrolled in the study. All associated medical records were retrospectively reviewed. Basic characteristics, diagnoses, age of surgical reconstruction, FB findings, associated anomalies, and survival durations were reviewed. Prognostic factors associated with the patients' mortality were analyzed.

Results: A total of 33 children were enrolled, and 91% of them were type C. The median age at the time of hospitalization was 26 days (range, birth to 9 years), including 20 (61%) low-birth-weight infants and 26 (79 %) referred patients. FB was performed in patients preoperatively (39%) and postoperatively (96.8%). Among them, 28 patients (85%) had associated anomalies, including 17 (52%) cardiac and 23 (70%) airway anomalies. The median age of 31 patients who underwent surgical reconstruction was 3 (range, 0–39) days. Esophageal anastomotic stricture (21/31, 67.7%) was the most common postsurgical complication. Twenty-three patients (74.2%) received postoperative FB-guided interventions, including balloon dilatation, laser therapy, and stent implantation. Among the 9 mortality cases, the median age at death was 270 (range, 4–3246) days. Significant factor associated with mortality was delayed (> 48 h old) or no surgical reconstruction (p = 0.030).

Conclusion: Delayed (>48-hour old) or no surgical reconstruction was significantly related to mortality in children with congenital EA and TEF. Preoperative and postoperative FB evaluations helped to facilitate diagnoses and nonsurgical managements and resolve the patients' tracheoesophageal problems.

Keywords: Children; Esophageal atresia; Fiberoptic bronchoscopy; prognosis; Tracheoesophageal fistula

1. INTRODUCTION

Esophageal atresia (EA), which is characterized by interruption of the esophagus, often occurs with or without tracheoesophageal fistula (TEF) and is a rare congenital malformation of the gastrointestinal tract. The overall incidence of EA and TEF occurs approximately 1 in 2500 to 1 in 5000 live births.[1–6]

The survival and life quality have improved in recent decades, owing to the advances in neonatal intensive critical care, anesthesia techniques, mechanical ventilation, nutritional support, antibiotic use, and surgical techniques.[1,2,7–10] The classification is based on the atresia of the esophagus and the presence of associated fistula to the trachea. Until now, the only curative option is surgical correction. Differentiating these anatomical variants is essential to facilitate the correct medical and surgical management.

More than half of these patients have additional congenital or chromosome malformation with syndromic presentation.[1,3,5,7] Patients with EA and TEF usually become symptomatic during the first 24 hours of life, with excessive salivation, choking, respiratory distress, and the inability of feeding. Usually, the first sign is failure to pass insert a catheter into the newborn infant's stomach. The diagnosis can be made by radiographic studies with water-soluble contrast, fiberoptic bronchoesophagoscopy (FB), or computed tomography.

Once the diagnosis is confirmed, the patient will need preoperative evaluations for aerodigestive anomalies and other associated congenital anomaly. The Waterston classification with regard to birth weight, pneumonia, and associated congenital anomalies was described initially in 1962.[11] Then Spitz classification, based on birth weight and the presence of major cardiac anomalies, was proposed in 1994 and was more widely used.[3,4,12] In 2018, Yamoto recommended the new classification, similar to the Spitz classification, using complex cardiac anomalies and birth weight.[13] Anastomotic leaks, esophageal anastomotic stricture (AS), gastroesophageal reflux (GER), and esophageal dysmotility are well-known, common, postoperative complications that occur in patients with EA and TEF.[3,4,14]

For many years, FB has been applied in children with airway problems for diagnostic and therapeutic purposes. In our institution, FB has been commonly employed to diagnose EA and TEF, determine associated respiratory anomalies, and provide interventions before surgical correction.[15–19] After surgery, FB would be used to evaluate and manage postoperative airway complications. Notwithstanding, using FB to assess EA and TEF and associated respiratory disorders is still not routinely applied in these patients preoperatively or postoperatively.[14,20–23]

The aim of the present study was to evaluate the prognostic factors and the role of preoperative and postoperative FB applications in managing children with EA and TEF.

2. METHODS

2.1 Patients

We retrospectively reviewed the medical records of hospitalized children who were diagnosed as EA and TEF between 2000 and 2017 at the Taipei Veterans General Hospital, which is a tertiary center. The inclusion criteria for the study were admission age of <18 years. The information extracted from the patients' charts included perioperative data (gender, gestational age, birth weight, diagnostic tool, and associated congenital anomalies), operative approach and findings, and postoperative data (morbidity, mortality, complications, and need for further intervention).

This study was approved by the Institutional Review Board (IRB) of Taipei Veterans General Hospital (IRB No. 2018-08-019AC).

2.2 Fiberoptic bronchoesophagoscopy procedures

FB examinations were performed by experienced pediatric pulmonologists in our department.[15–17,19] The noninvasive ventilation technique, using nasopharyngeal oxygen with intermittent nose closure and abdominal compression for maintaining airway, oxygenation, and ventilation, was applied throughout all of the FB procedures (Fig. 1).[18] Intravenous midazolam (0.5–1.0 mg/kg) and ketamine (1.0–2.0 mg/kg) were administered as sedative and analgesic agents, and a topical anesthetic (2% lidocaine, 0.2–0.5 ml/kg) was instilled into the upper airway via both nostrils as well as into the esophagus. These patients were heavily sedated with preservation of spontaneous breathing. When patients required balloon dilatation for esophageal anastomotic stricture, an angioplasty balloon catheter was orally inserted into the esophagus, followed by insertion of the FB behind the balloon segment (Fig. 2A). The appropriate balloon size was selected according to the location and the dimension of the stenotic portion. Laser therapy was performed for esophageal AS in a similar way. The laser fiber was inserted nasally via the inner channel of the endoscope into the lesion sites, and then fired to ablate the stenotic portion. An FB-guided stent implantation with uncovered balloon expandable metal stent was performed for symptomatic tracheomalacia, bronchostenosis, or refractory esophageal AS (Fig. 2B). All these FB findings and interventional procedures were recorded and analyzed.

2.3 Definition of cardiac anomalies and postoperative complications

Congenital heart disease was determined as a clinically identified cardiac defect other than patent ductus arteriosus and patent foramen ovale. Furthermore, complex cardiac anomalies were defined as cyanotic heart disease, such as right ventricular outflow tract stenosis, malformations leading to pulmonary artery and pulmonary vein stenosis, arterial vascular flow-dependent diseases, heart failure, and other complicated cardiac malformations, which required early operation with a cardiopulmonary bypass during early infancy. Tracheomalacia was defined as at least a 50% reduction of tracheal lumen with spontaneous quiet breathing before or after surgical correction. Anastomotic leaks were confirmed by clinical appearance after surgery. The presence of recurrent TEF was verified by postoperative FB with methyl blue stain. Esophageal AS was defined as narrowing at the level of the esophageal anastomosis with significant functional impairment and symptoms of which active treatment was indicated. GER was defined as reflux of gastric contents causing symptoms such as recurrent regurgitation with or without vomiting accompanied with poor weight gain, irritability, heartburn, or coughing.

2.4 Statistical evaluation

Statistical analysis was performed using the software SPSS (version 24, IBM corporation, Armonk, NY). The data are expressed as mean ± SD values and median with range or percentage where appropriate. The chi-squared or Fisher's exact tests were used to assess the differences between the categorical variables. The survival rate comparisons were analyzed using the Kaplan–Meier method, and significance was calculated using the log-rank test. The discrimination of different prognostic classification was compared by the area under the receiver operating characteristic (ROC) curve (AUC). Two-tailed p values of <0.05 were considered to be statistically significant for all the analyzes.

3. RESULTS

3.1 Characteristics of patients

During the 17-year study period, a total of 33 children with EA and TEF were enrolled in the analysis. One-third (n = 11) of them were admitted from 2000 to 2007, and the remaining two-thirds (n = 22) of the patients were admitted from 2008 to 2017. Seven (21.2%) infants were inborn patients. The remaining 26 (78.8%) infants were transferred from another hospital, and 17 of them received surgery before their transfer. The median age at the time of admission age was 26 days, and 21 (63.6%) infants were admitted at age <90 days.

The characteristics and clinical data are summarized in Table 1. There were slightly more females than males. Most of the infants were born with low birth weight (n = 20, 60.6%). The mean birth weight was 2404 ± 596 g (range, 770–3670 g). Regarding gross classification, the most common type was type C (n = 30, 90.9%). A total of 31 infants received surgical reconstruction, and the mean age of reconstruction was 5 ± 7 days (Table 1). Most of the infants required staged repair (58.1%) (Table 1).

3.2 Associated anomalies and preoperative FB examinations

Of the enrolled patients, associated anomalies were found in 28 (84.8%) patients (Table 2). In our study, airway and lung anomalies were the most frequent and occurred in 23 (69.7%) patients. Cardiac anomalies were detected in 17 (51.5%) patients, including seven patients with complex cardiac anomalies. Other associated anomalies included anorectal, genitourinary, extremities, vertebral, and chromosomal anomalies. A total of 10 patients
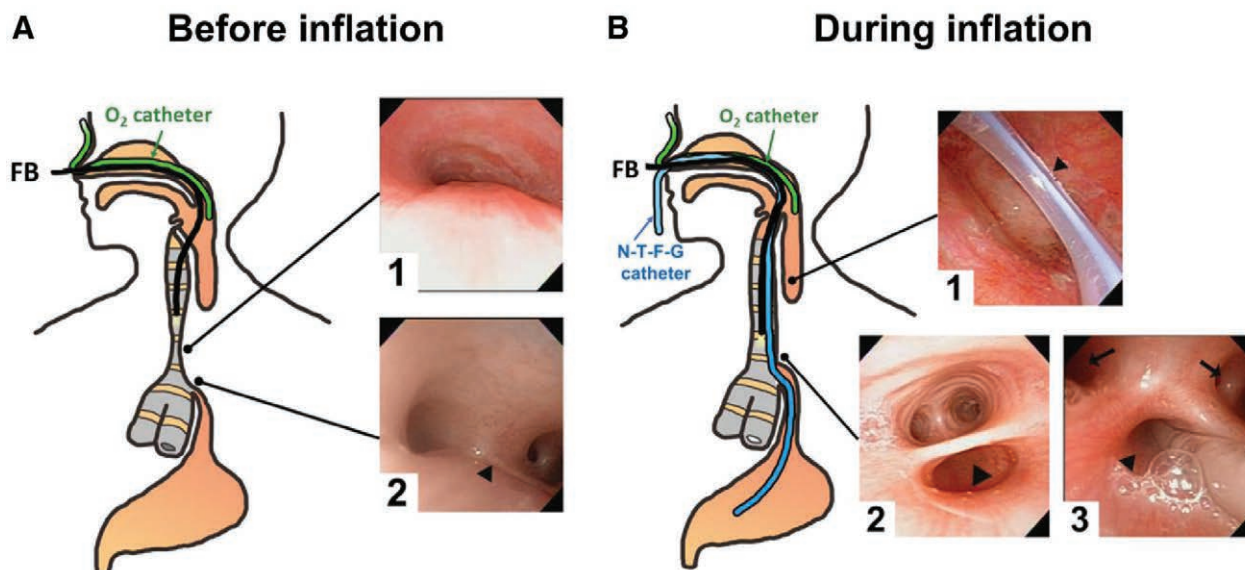
Fig. 1. Preoperative fiberoptic bronchoesophagoscopy (FB) in a patient with esophageal atresia and tracheoesophageal fistula combined with tracheomalacia. Before inflation (A), tracheomalacia (A-1) above the fistula (A-2) is presented. During inflation (B), rotation of catheter at esophageal pouch is presented (B-1). A nasal–tracheal–fistula–gastric tube through the fistula (B-2, arrowhead) is inserted for gastric decompression (B-3, arrowhead). Arrow indicates the left and right bronchus (B-3).
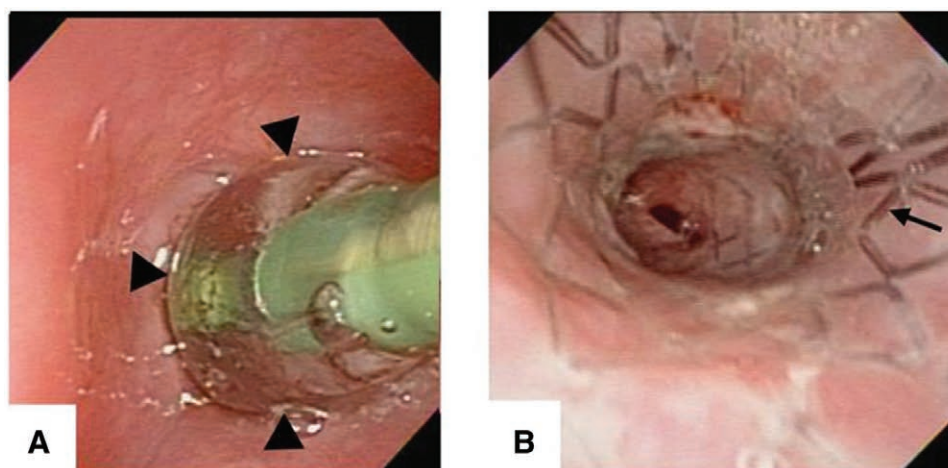

Fig. 2. Fiberoptic bronchoesophagoscopy-guided balloon dilatation (arrowhead) for esophageal anastomotic stricture (A) and tracheal stent implantation (arrow) for severe tracheomalacia (B).

(30.3%) fit the diagnostic criteria of VACTERL association of congenital anomaly (Table 2).

Among all 33 patients, there was one patient who did not receive any FB examination throughout their hospitalizations. Before surgery, 13 patients received FB examinations, including three patients with FB-guided endotracheal intubations and one patients with a nasal–tracheal–fistula–gastric catheter insertion for gastric juice drainage and gastric decompression to prevent aspiration before surgery (Fig. 1B).

3.3 Postsurgical complications and managements

After surgery (n = 31), most of the patients (n = 30, 96.8%) received FB examinations, and 23 patients required FB-guided interventions (Table 3).

Among the postsurgical patients, esophageal AS was the most common complication (n = 21, 67.7%), which was quickly diagnosed via FB examinations. Eighteen (58.1%) patients required FB-guided intervention to relieve the severity of esophageal AS; and three patients of them received surgery again because of both esophageal AS and recurrent TEF. Balloon dilatation was the most common procedure (n = 17). Additionally, seven patients received laser therapy, and two patients received stent implantations. Nine of them required more than one type of intervention to alleviate esophageal stenotic severity (Table 3).

The second common complication was GER (n = 21, 67.7%), and seven patients required fundoplication (Table 3). Anastomotic leak and recurrent TEF were also found in several patients.

Additionally, eight patients were coincidentally found to have tracheomalacia (n = 7) and bronchostenosis (n = 3) after FB examinations and required stent implantation (Table 3).

Table 1

Characteristics of 33 children with EA and TEF

Characteristics	Data
Gender, n (male/female)	15/18
Birth weight (mean ± SD)	2404 ± 596 g (range:770~3670 g)
Birth weight ≤ 2500g, n (%)	20 (60.6)
Birth weight ≤ 1500g, n (%)	2 (6.1)
Gestational age at birth, median (range) (weeks)	37 (27~41)
Born in our hospital, n (%)	7 (21.2)[a]
Transferred from other hospital, n (%)	26 (78.8)
Before surgery, n	9
After surgery, n	17
Age at admission, median (range) (days old)	26 (born ~ 3606)
Gross classification	
Type A, n (%)	2 (6.1)
Type C, n (%)	30 (90.9)
Unclassifiable, n (%)	1 (0.03)
Age of surgical reconstruction, median(range) (days)	3 (0–39)
Surgical reconstruction, n/n (%)	31/33 (93.9)
Performed at other/our hospital, n	17/14
Primary open anastomosis, n/n (%)	13/31 (41.9)
Staged repair, n/n (%)	18/31 (58.1)
Year of admission	
2000–2007, n (%)	11 (33.3)
Mortality, n (%)	4 (36.4)[a]
2008–2017, n (%)	22 (66.7)
Mortality, n (%)	5 (22.7)
Age at death, median (range) (days old)	270 (4–3246)
Overall survival rate, n/n (%)	24/33 (72.7)
Survival rate after reconstruction, n/n (%)	24/31 (77.4)

[a]Two mortality cases did not receive surgical reconstruction due to parents' refusal.

EA = esophageal atresia; TEF = tracheoesophageal fistula.

Table 2

Associated anomalies among 33 children with EA and TEF

Anomalies	n (%)
Any anomaly	28 (84.8)
Airway and lung	23 (69.7)
Tracheomlacia	21
Bronchostenosis	6
Lung hypoplasia	2
Laryngeal cleft	2
Cardiac	17 (51.5)
Ventricular septal defect	7
Tetralogy of the Fallot	5
Double outlet right ventricle	2
Total anomalous pulmonary venous return	1
Pulmonary stenosis	5
Coarctation of the aorta	1
Patent ductus arteriosus	2
Pulmonary hypertension	1
Anorectal	6 (18.2)
Genitourinary	5 (15.2)
Extremities	3 (9.1)
Vertebral	2 (6.1)
Chromosomal	2 (6.1)
VACTERL association	10 (30.3)

EA = esophageal atresia; TEF = tracheoesophageal fistula; VACTERL = vertebral defects, anal atresia, cardiac defects, tracheo-esophageal fistula, renal anomalies, and limb abnormalities.

3.4 Mortality and associated factor analysis

Nine patients died during the study period. Two of them did not receive surgery due to their parents' refusal. Other mortality

Table 3

Complications and therapeutic managements after surgery, and the use of FB in enrolled children with EA and TEF

	n	%
Never received FB among all 33 enrolled cases	1	3.0
Received surgical reconstruction	31	93.9
Complications among 31 post-surgical cases		
Esophageal AS	21[b]	67.7
Gastroesophageal reflux	21	67.7
Requiring fundoplication	7	
Anastomotic leak	10	32.2
Recurrent TEF	8	25.8
FB applications[a]		
Preoperative FB among all 33 enrolled cases	13	39.4
Diagnosis only	9	69.2
Diagnosis plus intervention	4	30.8
FB-guided ET intubation	3	
FB-guided NTFG tube intubation	1	
Postoperative FB among 31 post-surgical cases	30	96.8
Examination without intervention	7	23.3
Examination with therapeutic interventions	23	76.7
For esophageal AS (FB-guided)	18[c]	60.0
Balloon dilatation only	8	
Balloon dilatation + laser therapy	7	
Balloon dilatation + stent implantation	1	
Balloon dilatation + laser therapy + stent implantation	1	
Laser therapy only	1	
For airways (FB-guided)	8	26.7
Stent implantation for tracheomlacia	5	
Stent implantation for bronchostenosis	1	
Stent implantation for tracheomalcia plus bronchostenosis	2	

[a]Eleven cases received both preoperative and postoperative FB examinations.

[b]Three cases received surgery again due to associated other postoperative complications.

[c]Three cases also had stent implantations for tracheomalacia or bronchostenosis.

AS = anastomotic stricture; EA = esophageal atresia; ET = endotracheal tube; FB = fiberoptic bronchoesophagoscopy; NTFG = nasal-tracheal-fistula-gastric; TEF = tracheoesophageal fistula.

cases included recurrent TEF in three patients, complex cardiac anomaly in two patients, intractable respiratory failure even after airway stent implantation in one patient, and lung hypoplasia with sudden death at the age of 10 years in one patient. The mortality rates were 36.4% in patients admitted from 2000 to 2007 and 22.7% in patients admitted from 2008 to 2017 (Table 1).

According to the ROC analysis, the cutoff value of the age of reconstruction that affected the mortality was the age of 2 days (AUC, 0.869; $p < 0.001$).

The results of potential factors that might be associated with patients' mortality are analyzed and presented in Table 4. As shown, a significantly higher mortality was found in patients who received reconstruction at age > 2 days or no surgery (odds ratio: 26.24, 95% CI: 1.37–502.65; $p = 0.030$).

3.5 Survival and prognostic classification

The overall survival rate of the enrolled patients was 72.7%, and the postsurgical survival rate was 77.4%. There was a tendency toward improved survival rates during the years from 2008 to 2017 compared with that of the period from 2000 to 2007, although there was no statistical significance between these two periods (Fig. 3A). The survival curve revealed a significant difference between patients who received surgical reconstruction early (≤ 2 days) and delayed (> 2 days) or no surgery ($p = 0.017$) (Fig. 3B).

As a result of the ROC curve, the Yamoto new classification was better than the Spitz classification for predicting prognosis

Table 4

Odds ratio analysis for potential factors related to mortality in 33 children with EA and TEF

Variable	Died (n = 9)[a]	Survived (n = 24)	OR	95% CI	*p*
Birth weight ≤ 1500g	1 (11.1)	1 (4.2)	2.88	0.16–51.54	0.473
Non-type C classification	1 (11.1)	1 (4.2)	2.88	0.16–51.54	0.473
Born at other hospital	6 (66.7)	22 (91.7)	0.18	0.02–1.35	0.096
Preoperative FB	6 (66.7)	7 (29.2)	4.86	0.94–25.09	0.059
Airway anomaly	5 (55.6)	18 (75.0)	0.42	0.08–2.08	0.286
Cardiac anomaly	7 (77.8)	10 (41.7)	4.90	0.84–28.73	0.078
Complex cardiac anomaly	4 (44.4)	3 (12.5)	5.60	0.94–33.43	0.059
Delayed (>48 h) or no reconstruction	9 (100)	10 (41.7)	26.24	1.37–502.65	0.030
Postoperative complication[a]					
Esophageal AS[a]	3 (42.9)	18 (75.0)	0.25	0.04–1.45	0.123
Gastroesophageal reflux[a]	5 (71.4)	16 (66.7)	1.25	0.2–7.92	0.813
Recurrent TEF[a]	3 (42.9)	5 (20.8)	2.85	0.47–17.11	0.252
Anastomotic leak[a]	4 (57.1)	6 (25.0)	4.00	0.69–23.23	0.123

Data are presented as n (%).

[a]Two patients who did not receive surgical correction were excluded, and both of them died. The total postoperative mortality case number was 7.

CI = confidence interval; EA = esophageal atresia; FB = fiberoptic bronchoscopy; OR = odds ratio; TEF = tracheoesophageal fistula.

according to AUC, but the results were not significant (AUC, 0.722 vs 0.694; *p* = 0.663).

4. DISCUSSION

Our results indicated that delayed age of reconstruction or no surgery was significantly associated with mortality in children with EA and TEF. The overall survival rate in our patients was 72.7%. FB helped to facilitate diagnosis and nonsurgical interventions.

According to the recent literature, the survival rate of patients with EA and TEF varied from approximately 70% to 95%.[1,7,8,10,12,24,25] The survival rate of our postoperative patients was 77.4%. Indeed, approximately four-fifths of enrolled patients were referred from another hospital, and more than half of these patients had received surgical intervention before being admitted to our hospital. We had performed FB-guided interventions to relieve the patients' airway narrowing or malacia,[15–19] so more than half of the referred patients who were admitted to our hospital due to failure from previous managements in their original hospitals required additional airway management. The disease severity of our enrolled patients was much higher than expected. Previously, Yang et al.[26] reported 15 patients with EA and TEF admitted from 1994 to 2003 to our hospital and who had a survival rate of 46.7%, and they demonstrated that the mortality was affected by associated life-threatening anomalies. In this report, we monitored these patients for a longer duration. Although we noted a tendency of higher proportion of complex cardiac anomaly in mortality cases, no significance could be demonstrated. Improvements in the management of complex cardiac and airway anomalies during the last decade may influence patient outcome and factors associated with mortality.

Several studies have surveyed the prognostic factors in EA and TEF, and the predictive factors have altered on the basis of the different period and institution.[11–13,24,25] There are also several classification systems that predict mortality in view of preoperative factors. In 1962, Waterston proposed mortality associated with birth weight, additional congenital anomalies, and pneumonia.[11] Decades later, Spitz reevaluated the prognostic classification based on low birth weight and cardiac anomalies in 1994.[12] According to the Spitz classification, a new classification was published by Yamoto et al.[13] in 2018. According to the above date and other literature reports,[24,25] the preoperative factors that affected mortality were body weight and associated
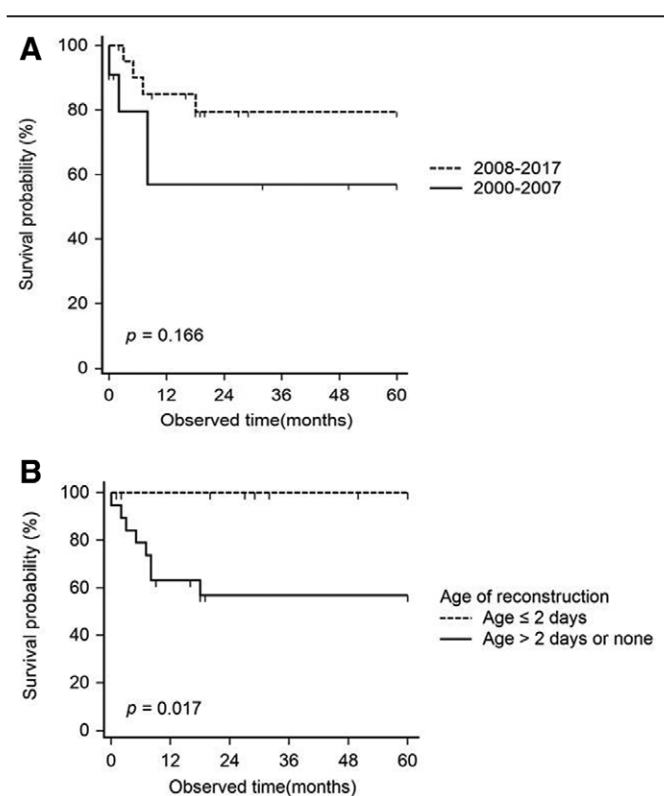

Fig. 3. Survival curve comparisons of enrolled patients (*p* > 0.05) admitted between 2000 and 2007 and 2008 and 2018 (A) and between surgical reconstruction at age ≤ 2 days and >2 days or no reconstruction (B).

congenital abnormalities. Furthermore, Masuya reported that associated cardiac and chromosomal anomalies affected the outcome of EA.[14]

Furthermore, we demonstrated that the timing of surgical reconstruction contributed to a patient's mortality. In the mortality cases, none of the patients had surgical reconstruction at age <48 hours. More than half of the survivors received surgical reconstruction within 48 hours of life (Table 4). The causes of delayed surgical correction might be due to a patient's illness or a surgeon's unavailability, so aerodigestive complications could

develop before the final surgical reconstruction. Our findings may provide clinical physicians with new information in planning therapeutic protocols for patients with congenital EA and TEF not to exceed 48 hours of life.

Our analysis demonstrated that >4/5 (84.8%) of enrolled patients with EA and TEF had associated anomalies. There is a large difference in the associated anomalies in percentage—varying widely from 46.6% to 81.9%—among the different literature reports or institutions.[1,27,28] It may likely be due to methodological differences, clinical definitions, inclusion/exclusion criteria, objective techniques, sample size, absence of autopsy, different diagnostic techniques, and missing data after transfer.[27] In addition, our results showed that the airway and pulmonary anomalies were higher than cardiac anomalies, which was different from the findings in other reports. Previous retrospective reports indicated that the incidence of respiratory problems in EA and TEF ranged from 24% to 79%.[29] The most possible reason is that we performed FB more in our patients compared with other studies so that we were able to demonstrate a higher proportion of airway anomalies that might not be easily identified clinically.

Using FB to manage patients with EA and TEF was increasing in the last decade.[5,9,19,22,30–32] We summarize publications on these cases with the use of preoperative and postoperative FB in Table 5. Among these reports, Atzori et al.[20] had reviewed their experience in 62 patients and concluded that FB enabled a better definition of anatomy and facilitated surgical repair. Deanovic et al.[34] reported their experience using FB-assisted repair for TEF and demonstrated how using FB facilitated the surgical outcome. In 2014, Tröbs and Finke[23] summarized their experience in 26 patients and suggested the routine use of rigid tracheobronchoscopy to identify the definite anatomy of TEF at the time of surgery. Sharma and Srinivas[36] also proved that laryngotracheobronchoscopy performed before reconstruction could be useful in the diagnosis and documentation of associated airway anomalies. Thus, tracheobronchoscopy before surgical repair of EA and TEF could characterize the anatomy, the number and site of fistula, and the length of the gap between proximal and distal esophagus and could assess associated anomalies such as tracheomalacia.[10,38–40] In addition, a

catheter placement or other tube cannulation via bronchoscopy was reported for continuous drainage out of gastric juice and gastric decompression to prevent aspiration pneumonia and gastrostomy.[41,42] In our patients, using FB did enable us to perform preoperative airway and esophagogastric evaluation as well as postoperative identification of AS. Additionally, we performed FB-guided insertion of an endotracheal tube or a nasal–tracheal–fistula–gastric tube for gastric drainage and decompression that prevented gastrostomy before complete surgical reconstruction. These patients tolerated the FB procedures well. Therefore, we think that FB is a safe technique when performed by professional and experienced physicians. We recommend that this procedure be performed before surgical reconstruction in cases of EA and TEF in order to adequately identify the aerodigestive anomalies and the potential of an interventional procedure to facilitate the preoperative stabilization of the patients.

After surgical intervention, AS, GER, and esophageal dysmotility are well-known common complications. Respiratory problems, such as residual tracheomalacia and bronchial stenosis, may also be attributed to GER, esophageal dysmotility, and the prolonged use of mechanical ventilation.[29] Previously, contrast esophagography was used to evaluate postoperative conditions.[25,31] Among the patients with AS, FB was performed for diagnosis and treatment and provided more reliable information than an esophagram.[43,44] Early routine screening was suggested; however, the timing of dilatation was controversial. Salö et al.[44] indicated that 39% of patients required esophageal dilatation after surgery, and they also concluded that the need for dilatation within 6 months postoperatively predicted the need for dilatation after 1 year. With regard to respiratory problems in the postoperative period, Thakkar et al.[22] reported that FB prior to reconstruction was not a reliable screening tool for symptomatic tracheomalacia. Therefore, repeating endoscopy in patients with symptoms was suggested after surgery. Although some published reports had shown that esophageal AS formation was commonly associated with recurrent lung infection,[45,46] our study did not find significant influence of postoperative AS on the mortality rate of our enrolled patients. The possible explanation may be due to our aggressive and early FB-guided

---

Table 5

Publications mentioned with the use of FB or bronchoscopy in children with EA and TEF

Author/year	Case no.	Study type	Survival (%)	Applications	
				Preoperative	**Postoperative**
Benjamin, 1981[33]	152	NM	NM	+	+
Yang et al., 2006[26]	15	Retrospective	46.7	+	+
Atzori et al., 2006[20]	62	Retrospective	NM	+	NM
Deanovic et al., 2007[34]	47	Prospective	NM	+	NM
Lal et al., 2013[35]	170	Questionnaire	NM	+ (60%)	-
Sfeir et al., 2013[30]	307	Prospective	95	+ (22%)	-
Tröbs et al., 2014[23]	26	Retrospective	NM	+	NM
Zani et al., 2014[31]	178	Questionnaire	NM	+ (43%)	-
Sharma and Srinivas, 2014[36]	88	Retrospective	NM	+	NM
Donoso et al., 2016[10]	129	Retrospective	94.6	+	+[a]
Lal et al., 2017[32]	396	Retrospective	92.7	+ (66%)	-
Rinkel et al., 2017[9]	12	NM	NM	+ (100%)	NM
Friedmacher et al., 2017[37]	109	Longitudinal cohort study	84.4	+	+
Porcaro et al., 2017[29]	105	Retrospective	NM	NM	+
Thakkar et al., 2018[22]	26	Prospective	NM	+ (88%)	+
Leibovitch et al., 2018[5]	69	Retrospective	81.2	NM	+
Chou et al., 2020	33	Retrospective	72.7	+ (39.4)	+

[a]Balloon dilatations were reported for ASs in 68 cases.

AS = anastomotic stricture; EA = esophageal atresia; FB = fiberoptic bronchoesophagoscopy; NM = not mentioned; TEF = tracheoesophageal fistula.

diagnosis and intervention to relief these patients' symptoms. Undoubtedly, postoperative complications and respiratory problems affected the quality of life and the children's growth. Thus, FB in the postoperative stage has an effective role in diagnosis and therapeutic managements.

There were several limitations in our study. The first limitation is that this is a single-center study and that most of the enrolled patients were referred from other hospitals due to the patients' difficult clinical situations. Therefore, the disease severity might be higher than expected, and there were wide variations in their management histories. Second, this is a retrospective study, and the results are based on chart records, which may have some potential results bias. Third, a long duration of study allowed us to compare the survival rates of different years but may have led to technologies and equipment of different time backgrounds that may attribute to bias.

In conclusion, delayed (> 48 hours old) or no surgical reconstruction significantly related to mortality in children with congenital EA and TEF. Preoperative and postoperative FB evaluations plus therapeutic interventions might have helped the diagnoses and nonsurgical managements to relieve patients' tracheoesophageal problems.

REFERENCES

1. Cassina M, Ruol M, Pertile R, Midrio P, Piffer S, Vicenzi V, et al. Prevalence, characteristics, and survival of children with esophageal atresia: a 32-year population-based study including 1,417,724 consecutive newborns. *Birth Defects Res A Clin Mol Teratol* 2016;**106**:542–8.
2. Pinheiro PF, Simões e Silva AC, Pereira RM. Current knowledge on esophageal atresia. *World J Gastroenterol* 2012;**18**:3662–72.
3. Spitz L. Esophageal atresia. Lessons I have learned in a 40-year experience. *J Pediatr Surg* 2006;**41**:1635–40.
4. Spitz L. Oesophageal atresia. *Orphanet J Rare Dis* 2007;**2**:24.
5. Leibovitch L, Zohar I, Maayan-Mazger A, Mazkereth R, Strauss T, Bilik R. Infants born with esophageal atresia with or without tracheo-esophageal fistula: short- and long-term outcomes. *Isr Med Assoc J* 2018;**20**:161–6.
6. Nomura A, Yamoto M, Fukumoto K, Takahashi T, Ohyama K, Sekioka A, et al. Evaluation of developmental prognosis for esophageal atresia with tracheoesophageal fistula. *Pediatr Surg Int* 2017;**33**:1091–5.
7. Wang B, Tashiro J, Allan BJ, Sola JE, Parikh PP, Hogan AR, et al. A nationwide analysis of clinical outcomes among newborns with esophageal atresia and tracheoesophageal fistulas in the United States. *J Surg Res* 2014;**190**:604–12.
8. Bakal U, Ersoz F, Eker I, Sarac M, Aydin M, Kazez A. Long-term prognosis of patients with esophageal atresia and/or tracheoesophageal fistula. *Indian J Pediatr* 2016;**83**:401–4.
9. Rinkel R, Van Poll D, Sibarani-Ponsen R, Sleeboom C, Bakx R. Bronchoscopy and fogarty balloon insertion of distal tracheo-oesophageal fistula for oesophageal atresia repair with video illustration. *Ann Otol Rhinol Laryngol* 2017;**126**:6–8.
10. Donoso F, Kassa AM, Gustafson E, Meurling S, Lilja HE. Outcome and management in infants with esophageal atresia — a single centre observational study. *J Pediatr Surg* 2016;**51**:1421–5.
11. Waterston DJ, Carter RE, Aberdeen E. Oesophageal atresia: tracheo-oesophageal fistula. A study of survival in 218 infants. *Lancet* 1962;**1**:819–22.
12. Spitz L, Kiely EM, Morecroft JA, Drake DP. Oesophageal atresia: at-risk groups for the 1990s. *J Pediatr Surg* 1994;**29**:723–5.
13. Yamoto M, Nomura A, Fukumoto K, Takahashi T, Nakaya K, Sekioka A, et al. New prognostic classification and managements in infants with esophageal atresia. *Pediatr Surg Int* 2018;**34**:1019–26.
14. Masuya R, Kaji T, Mukai M, Nakame K, Kawano T, Machigashira S, et al. Predictive factors affecting the prognosis and late complications of 73 consecutive cases of esophageal atresia at 2 centers. *Pediatr Surg Int* 2018;**34**:1027–33.
15. Hsieh KH, Chou YL, Soong WJ, Lee YS, Tsao PC. Long-term management and outcomes of tracheobronchial stent by flexible bronchoscopy in infants <5 kg: a 13-year single-center experience. *J Chin Med Assoc* 2019;**82**:727–31.
16. Lin YT, Lee YS, Jeng MJ, Chen WY, Tsao PC, Chan IC, et al. Flexible bronchoscopic findings and the relationship to repeated extubation failure in critical children. *J Chin Med Assoc* 2018;**81**:804–10.
17. Peng YY, Soong WJ, Lee YS, Tsao PC, Yang CF, Jeng MJ. Flexible bronchoscopy as a valuable diagnostic and therapeutic tool in pediatric intensive care patients: a report on 5 years of experience. *Pediatr Pulmonol* 2011;**46**:1031–7.
18. Soong WJ, Jeng MJ, Lee YS, Tsao PC, Harloff M, Matthew Soong YH. A novel technique of non-invasive ventilation: pharyngeal oxygen with nose-closure and abdominal-compression–aid for pediatric flexible bronchoscopy. *Pediatr Pulmonol* 2015;**50**:568–75.
19. Soong WJ, Tsao PC, Lee YS, Yang CF. Flexible endoscopy for pediatric tracheobronchial metallic stent placement, maintenance and long-term outcomes. *PLoS One* 2018;**13**:e0192557.
20. Atzori P, Iacobelli BD, Bottero S, Spirydakis J, Laviani R, Trucchi A, et al. Preoperative tracheobronchoscopy in newborns with esophageal atresia: does it matter? *J Pediatr Surg* 2006;**41**:1054–7.
21. Tambucci R, Angelino G, De Angelis P, Torroni F, Caldaro T, Balassone V, et al. Anastomotic strictures after esophageal atresia repair: incidence, investigations, and management, including treatment of refractory and recurrent strictures. *Front Pediatr* 2017;**5**:120.
22. Thakkar H, Upadhyaya M, Yardley IE. Bronchoscopy as a screening tool for symptomatic tracheomalacia in oesophageal atresia. *J Pediatr Surg* 2018;**53**:227–9.
23. Tröbs R-B, Finke W. Endoscopic detection and surgical repair of congenital tracheo-esophageal-fistula (TEF) ± esophageal atresia (EA). *Open Journal of Pediatrics* 2014;**4**:283.
24. Peters RT, Ragab H, Columb MO, Bruce J, MacKinnon RJ, Craigie RJ. Mortality and morbidity in oesophageal atresia. *Pediatr Surg Int* 2017;**33**:989–94.
25. Ammar S, Sellami S, Sellami I, A BH, Jarraya A, Zouari M, et al. Management of esophageal atresia and early predictive factors of mortality and morbidity in a developing country. *Dis Esophagus* 2019;**32**.
26. Yang CF, Soong WJ, Jeng MJ, Chen SJ, Lee YS, Tsao PC, et al. Esophageal atresia with tracheoesophageal fistula: ten years of experience in an institute. *J Chin Med Assoc* 2006;**69**:317–21.
27. Stoll C, Alembik Y, Dott B, Roth MP. Associated anomalies in cases with esophageal atresia. *Am J Med Genet A* 2017;**173**:2139–57.
28. Forrester MB, Merz RD. Epidemiology of oesophageal atresia and tracheo-oesophageal fistula in Hawaii, 1986-2000. *Public Health* 2005;**119**:483–8.
29. Porcaro F, Valfré L, Aufiero LR, Dall'Oglio L, De Angelis P, Villani A, et al. Respiratory problems in children with esophageal atresia and tracheoesophageal fistula. *Ital J Pediatr* 2017;**43**:77.
30. Sfeir R, Bonnard A, Khen-Dunlop N, Auber F, Gelas T, Michaud L, et al. Esophageal atresia: data from a national cohort. *J Pediatr Surg* 2013;**48**:1664–9.
31. Zani A, Eaton S, Hoellwarth ME, Puri P, Tovar J, Fasching G, et al. International survey on the management of esophageal atresia. *Eur J Pediatr Surg* 2014;**24**:3–8.
32. Lal DR, Gadepalli SK, Downard CD, Ostlie DJ, Minneci PC, Swedler RM, et al.; Midwest Pediatric Surgery Consortium. Perioperative management and outcomes of esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg* 2017;**52**:1245–51.
33. Benjamin B. Endoscopy in esophageal atresia and tracheoesophageal fistula. *Ann Otol Rhinol Laryngol* 1981;**90**(4 Pt 1):376–82.
34. Deanovic D, Gerber AC, Dodge-Khatami A, Dillier CM, Meuli M, Weiss M. Tracheoscopy assisted repair of tracheo-esophageal fistula (TARTEF): a 10-year experience. *Paediatr Anaesth* 2007;**17**:557–62.
35. Lal D, Miyano G, Juang D, Sharp NE, St Peter SD. Current patterns of practice and technique in the repair of esophageal atresia and tracheoesophageal fistua: an IPEG survey. *J Laparoendosc Adv Surg Tech A* 2013;**23**:635–8.
36. Sharma N, Srinivas M. Laryngotracheobronchoscopy prior to esophageal atresia and tracheoesophageal fistula repair–its use and importance. *J Pediatr Surg* 2014;**49**:367–9.

37. Friedmacher F, Kroneis B, Huber-Zeyringer A, Schober P, Till H, Sauer H, et al. Postoperative complications and functional outcome after esophageal atresia repair: results from longitudinal single-center follow-up. *J Gastrointest Surg* 2017;**21**:927–35.

38. Taghavi K, Stringer MD. Preoperative laryngotracheobronchoscopy in infants with esophageal atresia: why is it not routine? *Pediatr Surg Int* 2018;**34**:3–7.

39. Higano NS, Bates AJ, Tkach JA, Fleck RJ, Lim FY, Woods JC, et al. Pre- and post-operative visualization of neonatal esophageal atresia/tracheoesophageal fistula via magnetic resonance imaging. *J Pediatr Surg Case Rep* 2018;**29**:5–8.

40. Parolini F, Boroni G, Stefini S, Agapiti C, Bazzana T, Alberti D. Role of preoperative tracheobronchoscopy in newborns with esophageal atresia: a review. *World J Gastrointest Endosc* 2014;**6**:482–7.

41. Papoff P, Cicchetti R, Montecchia F, Midulla F, Ceccanti S, Cozzi D. Intentional tracheoesophageal fistula cannulation for gastric decompression in type C esophageal atresia. *Paediatr Anaesth* 2018;**28**:367–9.

42. Pepper VK, Boomer LA, Thung AK, Grischkan JM, Diefenbach KA. Routine bronchoscopy and fogarty catheter occlusion of tracheoesophageal fistulas. *J Laparoendosc Adv Surg Tech A* 2017;**27**:97–100.

43. Krishnan U, Mousa H, Dall'Oglio L, Homaira N, Rosen R, Faure C, et al. ESPGHAN-NASPGHAN guidelines for the evaluation and treatment of gastrointestinal and nutritional complications in children with esophageal atresia-tracheoesophageal fistula. *J Pediatr Gastroenterol Nutr* 2016;**63**:550–70.

44. Salö M, Stenström P, Anderberg M, Arnbjörnsson E. Anastomotic strictures after esophageal atresia repair: timing of dilatation during the first two postoperative years. *Surg J (N Y)* 2018;**4**:e62–5.

45. Shah R, Varjavandi V, Krishnan U. Predictive factors for complications in children with esophageal atresia and tracheoesophageal fistula. *Dis Esophagus* 2015;**28**:216–23.

46. Kovesi T. Aspiration risk and respiratory complications in patients with esophageal atresia. *Front Pediatr* 2017;**5**:62.