



Original Article



Comparative Analysis of Two National Surveys on Esophageal Atresia With or Without Tracheoesophageal Fistula: A Retrospective Study by the Korean Association of Pediatric Surgeons

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Conflict of Interest

No potential conflict of interest relevant to this article was reported.

ABSTRACT

Purpose: The Korean Association of Pediatric Surgeons (KAPS) conducts annual nationwide surveys on various aspects of pediatric surgical diseases, with the results being discussed during KAPS's annual spring meetings.

Methods: KAPS conducted two national surveys, in 1995 and 2016, to investigate esophageal atresia (EA) with or without tracheoesophageal fistula (TEF). The authors analyzed data from these surveys to identify differences or changes in the annual occurrence, demographic characteristics, clinical presentation, preoperative diagnostic methods, anatomical type, associated anomalies, surgical treatment, and postoperative outcomes among patients with EA/TEF treated by KAPS members.

Results: The first and second national surveys included 148 and 211 patients with EA/TEF, respectively. Excessive salivation was the most prevalent clinical symptom in both surveys. Type C was the most common form of EA/TEF in both surveys. The first survey included 126 patients, all of whom underwent open surgery. In the second survey, 152 (78.4%) of 194 patients underwent open surgery, while 34 (17.5%) underwent thoracoscopic surgery. Primary esophageal repair was performed on 96 (76.2%) of 126 patients in the first survey and on 160 (82.5%) of 194 patients in the second survey. Anastomotic strictures developed in 21.4% and 32.5% of patients, anastomotic leakage in 22.2% and 10.3%, recurrent fistula in 2.4% and 4.2% during the first and second surveys, respectively. The respective survival rates for group A were 90.2% and 98.3% in the first and second surveys. For group B, the rates were 73.9% and 98.1%, and for group C, they were 34.5% and 68.1%, respectively, according to the Waterston classifications.

Conclusion: These nationwide surveys provide comprehensive information on the status, detailed treatment, and outcomes for Korean pediatric patients with EA/TEF. They are anticipated to be an invaluable resource and guide for pediatric surgeons seeking to expand their knowledge on EA/TEF and its treatment options.

Keywords: Esophageal atresia; Tracheoesophageal fistula; Children; Survey

Author Contributions

Conceptualization: P.J., L.N.H.; Data curation: P.J., L.N.H.; Formal analysis: P.J., L.N.H.; Investigation: P.J., L.N.H.; Methodology: P.J., L.N.H.; Resources: P.J., L.N.H.; Validation: P.J., L.N.H.; Writing - original draft: P.J.; Writing - review & editing: L.N.H.

INTRODUCTION

Esophageal atresia (EA) with or without tracheoesophageal fistula (TEF) is a congenital disease of the esophagus, occurring in 1 in 3,000–4,500 live births [1]. The exact cause of EA/TEF remains uncertain, though it is thought to result from a combination of environmental and genetic factors. Signs and symptoms of EA/TEF can manifest shortly after birth, including excessive salivation, swallowing difficulties, choking, coughing, and respiratory distress. EA/TEF is often associated with congenital anomalies and disorders, leading to significant morbidity and mortality [2]. Owing to advancements facilitating prompt diagnosis and appropriate surgical interventions, the survival rate for individuals with EA/TEF has significantly improved over time. However, there may be ongoing concerns and complications requiring continuous medical attention. A multidisciplinary team, including pediatric surgeons, neonatologists, and other medical experts, often provides comprehensive care for infants with EA/TEF. Since 1991, the Korean Association of Pediatric Surgeons (KAPS) has conducted annual nationwide surveys on various topics related to pediatric surgical diseases. The findings of these surveys are analyzed at KAPS's annual spring meetings. KAPS has conducted two nationwide surveys, in 1995 and 2016, on EA/TEF. The results of these surveys were discussed at the 11th and 32nd annual spring KAPS meetings, held on June 16, 1995, in Seoul, and on June 6, 2016, at the WE hotel in Jeju.

METHODS

Over 3 years, from 1992 through 1994, 28 KAPS members working at 23 pediatric surgical centers in Korea participated in the first national KAPS survey on EA/TEF. The results were published in the *Journal of the Korean Association of Pediatric Surgeons* in 1995 [1]. To identify any differences or changes in annual incidence, demographic characteristics, clinical presentation, preoperative diagnostic methods, anatomical types, anomalies, surgical treatment, and postoperative outcomes associated with EA/TEF, a second national KAPS EA/TEF survey was conducted. This second survey took place over 4 years, from 2012 through 2015. Prior to conducting the survey, we created a case registration form designed to capture information on patient age, patient sex, clinical presentation, preoperative diagnostic workup, type of EA/TEF, surgical treatment, and outcomes. KAPS members filled out the case registration form, which was sent to 19 pediatric surgical centers, encompassing a total of 211 patients. Differences in the case registration forms between the first and second national surveys posed challenges for direct data comparison. Nonetheless, the findings from both surveys were compared without any statistical validation. The study protocol was approved by the Institutional Review Board of Kyungpook National University Hospital (IRB No. 2024-02-027).

RESULTS

1. Registered case number and incidence by year

In the first national survey, 148 patients were included, comprising 39, 43, and 66 patients in 1992, 1993, and 1994, respectively. The second national survey included a total of 211 patients, with counts of 60, 61, 46, and 44 patients in 2012, 2013, 2014, and 2015, respectively. The respective annual EA/TEF incidence rates determined by the surveys were 1 in 14,385 and 1 in 8,508 (Table 1).

Table 1. Registered case number and incidence by year

1st national survey (n=148)			2nd national survey (n=211)		
Year	No. of cases/Birth No.	Incidence	Year	No. of cases/Birth No.	Incidence
1992	39/723,927	1/18,562	2012	60/484,550	1/8,076
1993	43/702,546	1/16,338	2013	61/436,455	1/7,155
1994	66/702,546	1/10,645	2014	46/435,435	1/9,466
			2015	44/438,700	1/9,970
Total	148/2,129,019	1/14,385	Total	211/1,795,140	1/8,508

2. Sex, gestational age, and body weight at birth

The first national survey included 69 males and 73 females, for a male-to-female ratio of 1:1.1. In the second national survey, the male-to-female ratio was 1.24:1, with 117 males and 94 females. The second national survey reported a gestational age range from 27 weeks and 3 days to 41 weeks and 5 days (mean, 37 weeks), along with a mean birth weight of 2,510 g (range, 665–4,000 g). Data on gestational age and body weight at birth in the first national survey were missing.

3. Clinical presentation

Polyhydramnios was observed in 46 (31.1%) and 77 (36.5%) pregnancies in the first and second national surveys, respectively. The most common symptom of EA/TEF was excessive salivation, reported for 98 (66.2%) patients in the first survey and 106 (50.2%) in the second survey. Other prevalent symptoms included respiratory difficulty in 62 (41.9%) and 93 (44.1%), cyanosis in 49 (33.1%) and 57 (27.0%), and choking after feeding in 36 (24.3%) and 30 (14.2%) of the cases, respectively. In the second national survey, 69 (32.7%) of the patients were diagnosed via prenatal evaluations (**Table 2**).

4. Preoperative diagnostic workup

KAPS members reported on preoperative diagnostic workup methods. Infantography with a radiopaque tube and renal ultrasonography were most frequently reported, having been performed on 128 (86.5%) and 51 (34.5%) patients, respectively, in the first survey. In the second survey, these numbers were 182 (86.3%) for infantography and 129 (61.1%) for ultrasonography. In the second national survey, 18 (8.5%) patients underwent chest computed tomography, a procedure not used during the period covering the first survey (**Table 3**).

5. Anatomical EA/TEF types

Type C was the most common anatomical type of EA/TEF in both surveys, with 126 (85.1%) and 179 (84.8%) patients, respectively, followed by type A with 8 (5.4%) and 20 (9.5%) patients, type D with 2 (1.4%) and 5 (2.4%) patients, type E with 3 (2.0%) and 4 (1.9%) patients, and type B with 1 (0.7%) and 1 (0.5%) patient (**Table 4**).

Table 2. Clinical presentation

Clinical presentation	1st national survey (n=148)	2nd national survey (n=211)
Maternal polyhydramnios	46 (31.1)	77 (36.5)
Excessive salivation	98 (66.2)	106 (50.2)
Respiratory difficulty	62 (41.9)	93 (44.1)
Cyanosis	49 (33.1)	57 (27.0)
Choking after feeding	36 (24.3)	30 (14.2)
Recurrent pneumonia	3 (2.0)	9 (4.3)
Gastric distention	17 (11.5)	15 (7.1)
Gastric perforation	-	3 (1.4)
Others	8 (5.4)	-

Values are presented as number of cases (%).

Table 3. Preoperative diagnostic workup

Preoperative diagnostic workup	1st national survey (n=148)	2nd national survey (n=211)
Infantography with a radiopaque tube	128 (86.5)	182 (86.3)
Infantography with contrast	40 (27.0)	7 (3.3)
Esophagography	7 (4.7)	25 (11.9)
Renal ultrasonography	51 (34.5)	129 (61.1)
Echocardiography	51 (34.5)	-
Chest computed tomography	-	18 (8.5)
Bronchoscopy	-	8 (3.8)
Others	5 (3.4)	21 (10.0)

Values are presented as number of cases (%).

6. Associated anomalies

The cardiovascular system was the most commonly involved in associated anomalies, with 46 (31.1%) and 98 (46.4%) patients, followed by the musculoskeletal system with 15 (10.1%) and 40 (19.0%) patients in the first and second national surveys, respectively (**Table 5**).

7. Operative procedure

In the first national survey, 126 of 148 patients—excluding 22 patients who died or withdrew before surgery—underwent open surgery. This group included 96 (76.2%) who underwent primary repair, 13 (10.3%) who underwent delayed primary repair, and 17 (13.5%) who underwent staged repair. In the second national survey, surgical treatment methods were analyzed for 194 of 211 patients, after excluding 9 patients who died before surgery and 8 with insufficient data. Open surgery, thoracoscopic surgery, and open conversion were performed on 152 (78.4%), 34 (17.5%), and 6 (3.1%) patients, respectively. Of these, 160 patients (82.5%) underwent primary repair, 26 (13.4%) underwent delayed primary repair, 4 (2.1%) underwent gastric pull-up, and 2 (1.0%) underwent gastric tube creation (**Table 6**).

Table 4. Anatomical type of EA with or without TEF

Anatomical type	1st national survey (n=148)	2nd national survey (n=211)
A (Pure EA)	8 (5.4)	20 (9.5)
B (EA with proximal TEF)	1 (0.7)	1 (0.5)
C (EA with distal TEF)	126 (85.1)	179 (84.8)
D (EA with double TEF)	2 (1.4)	5 (2.4)
E (H-type TEF)	3 (2.0)	4 (1.9)
Others	8 (5.4)	2 (0.9)

Values are presented as number of cases (%).

EA, esophageal atresia; TEF, tracheoesophageal fistula

Table 5. Associated anomalies

Associated anomalies	1st national survey (n=148)	2nd national survey (n=211)
Cardiovascular system	46 (31.1)	98 (46.4)
Gastrointestinal system	7 (4.7)	41 (19.4)
Musculoskeletal system	15 (10.1)	40 (19.0)
Genitourinary system	10 (6.8)	38 (18.0)
Chromosomal anomalies	-	5 (2.4)
VACTERL	-	34 (16.1)
Central nerve system	6 (4.1)	-
Face	6 (4.1)	-
Others	7 (4.7)	-
None	-	66 (31.3)

Values are presented as number of cases (%).

VACTERL, vertebral defects, anorectal malformations, cardiac defects, tracheoesophageal fistula with or without oesophageal atresia, renal malformations, and limb defects.

8. Postoperative complications

Postoperative complications included anastomotic strictures in 27 (21.4%) and 63 (32.5%) patients, anastomotic leakage in 28 (22.2%) and 20 (10.3%) patients, recurrent fistula in 3 (2.4%) and 8 (4.2%) patients, and respiratory complications in 65 (51.6%) and 33 (17.0%) patients from the first and second national surveys, respectively (**Table 7**).

9. Postoperative course

In the first and second national surveys, 56 (74.6%) and 107 (55.2%) patients were reported to be doing well, while 6 (8%) and 36 (18.6%) patients were doing well but with postoperative gastroesophageal reflux disease. Four (5.3%) and 22 (11.3%) patients developed esophageal strictures requiring dilatation (**Table 8**).

10. Survival rate according to the Waterston classification

The survival rates, according to the Waterston classification, were 90.2% and 98.3% in group A, 73.9% and 98.1% in group B, and 34.5% and 68.1% in group C for the first and second national surveys, respectively (**Table 9**).

Table 6. Operative procedure in the second national survey

Operative procedure	Type A (n=20)	Type B (n=1)	Type C (n=164)	Type D (n=5)	Type E (n=4)	Total (n=194)
Mode of surgery						
Open	15	1	129	5	2	152 (78.4%)
Thoracoscopic	3		31			34 (17.5%)
Conversion	2		4			6 (3.1%)
Unknown					2	2 (1.0%)
Operative procedure						
Primary repair	2		151	5	2	160 (82.5%)
Delayed primary repair	15	1	10			26 (13.4%)
Gastric tube	1		1			2 (1.0%)
Gastric pull-up	2		2			4 (2.1%)
Unknown					2	2 (1.0%)

Table 7. Postoperative complications

Postoperative complications	1st national survey (n=126)	2nd national survey (n=194)
Anastomotic stricture	27 (21.4)	63 (32.5)
Anastomotic leakage	28 (22.2)	20 (10.3)
Recurrent fistula	3 (2.4)	8 (4.2)
Respiratory complications	65 (51.6)	33 (17.0)
Gastroesophageal reflux	4 (3.2)	37 (19.1)
Wound infection	4 (3.2)	5 (2.6)
Bleeding requiring transfusion	-	2 (1.0)
Sepsis	23 (18.3)	-
Others	2 (1.6)	11 (5.7)

Values are presented as number of cases (%).

Table 8. Postoperative course

Postoperative course	1st national survey (n=75)	2nd national survey (n=194)
Doing well	56 (74.6)	107 (55.2)
Doing well but GER	6 (8.0)	36 (18.6)
Recurrent respiratory infection	9 (12.0)	15 (7.7)
Esophageal stricture with dilatation	4 (5.3)	22 (11.3)
Death	-	10 (5.2)
Unknown	-	4 (2.1)

Values are presented as number of cases (%).
GER, gastroesophageal reflux.

Table 9. Survival rate according to the Waterston classification

Waterston group	1st national survey (n=126)	2nd national survey (n=211)
	Survivor/No. of cases (%)	Survivor/No. of cases (%)
A	46/51 (90.2)	59/60 (98.3)
B	34/46 (73.9)	102/104 (98.1)
C	10/29 (34.5)	32/47 (68.1)
Total	90/126 (71.4)	192/211 (91.0)

A: Birth weight over 2,500g and well; B: Birth weight 1,800g to 2,500g and well or over 2,500g with moderate pneumonia and congenital anomaly; C: Birth weight under 1,800g and well or 1,800g to 2,500g with severe pneumonia and congenital anomaly.

Table 10. Comparison between open and thoracoscopic surgery in the second national survey

Variables	Open surgery (n=150)	Thoracoscopic surgery (n=34)
Operation time (min)	181.1±73.9	182.5±94.7
Complication		
Anastomotic stricture	45 (30.0)	11 (32.4)
Anastomotic leakage	10 (6.7)	8 (23.5)
Recurrent fistula	6 (4.0)	2 (5.9)
Gastroesophageal reflux	30 (20.0)	7 (20.6)
Pneumonia	38 (25.3)	1 (2.9)
Wound infection	5 (3.3)	0 (0.0)
Mortality	6 (4.0)	2 (5.9)

Values are presented as mean ± standard deviation or number of cases (%).

11. Comparison between open and thoracoscopic surgery in the second national survey

For both open and thoracoscopic operations, the mean operation times were 181.1±73.9 and 182.5±94.9 minutes, respectively. Postoperative complications following open and thoracoscopic procedures anastomotic strictures in 45 (30%) and 11 (32.4%) patients, anastomotic leakage in 10 (6.7%) and 8 (23.5%) patients, recurrent fistulas in 6 (4%) and 2 (5.9%) patients, gastroesophageal reflux in 30 (20%) and 7 (20.6%) patients, pneumonia in 38 (25.3%) patients and 1 (2.9%) patient, and wound infections in 5 (3.3%) and 0 (0%) patients, respectively. Six patients (4%) died following open surgery and two patients (5.9%) following thoracoscopic surgery (**Table 10**).

DISCUSSION

Twenty-one years after conducting the first national survey on EA/TEF, KAPS carried out the second national survey. The comparison of the two national surveys revealed that while the incidence of EA/TEF was slightly lower in the second survey, the demographics of patients, clinical presentation, preoperative diagnostic methods, anatomical types, and associated anomalies remained largely unchanged. However, in the first national survey, all patients underwent open surgery; in contrast, in the second national survey, 78.4% underwent open surgery, and 17.5% received thoracoscopic surgery.

Numerous multicenter investigations on the treatment outcomes of EA/TEF have been conducted and published since KAPS's first national survey in 1995 [2-5]. Keefe et al. [2] reported an in-hospital mortality rate of 12.7%. Lower birth weight and the presence of concurrent congenital abnormalities were significantly associated with higher mortality rates and longer hospital stays. According to the Midwest Pediatric Surgery Consortium, patients who received a postoperative transanastomotic tube experienced increased stricture rates,

and those with prosthetic material placed between the esophageal and tracheal suture lines encountered higher leakage rates. They also found no benefit of prolonged acid suppression medication or of the use of prophylactic perioperative antibiotics for more than 24 hours after EA/TEF repair [4].

In the past, thoracotomies were performed to treat EA/TEF, but thorascopies have gained popularity in recent years. Thoracoscopy offers advantages such as improved visibility, reduced chest wall deformation, and favorable cosmesis, though its challenges include potential hypercapnia or acidosis during surgery, technical difficulties, a lengthy learning curve, and longer operating times [3]. While considerable research has evaluated the treatment outcomes of thoracoscopy, most studies have been small retrospective reviews or single-institutional cohort studies. Consequently, there are concerns about bias in surgeons' patient selection for thorascopic surgery. Therefore, many systematic reviews and meta-analyses have recently been undertaken to compare the efficacy of thoracotomy and thoracoscopy as treatment methods [6-9].

In a study by Yang et al. [6], the rates of leakage and strictures following EA/TEF repair were comparable between thorascopic and conventional open repair techniques. Thorascopic repair offered benefits such as shorter hospital stays, earlier extubation times, and the initiation of oral feeding, despite longer operating times [6]. Additionally, Way et al. [7] demonstrated that thorascopic repair for EA/TEF is safe and does not result in higher morbidity compared with the open technique. While thorascopic repair required longer operating times, it had comparable rates of complications, anastomotic leakage, strictures, and fundoplication requirements to open repair [7]. Wu et al. [9] found that thorascopic repair could shorten the hospital stay and the time to first oral feeding but was associated with longer operative times compared with open repair. There was no significant difference in the frequency of leakage, strictures, pulmonary complications, fundoplication for gastroesophageal reflux disease, or blood loss between the groups undergoing open and thorascopic repairs [9]. On the other hand, thorascopic EA/TEF repair by Thakkar et al. [10] was associated with a higher stricture rate than open surgery. In the second national KAPS survey, anastomotic leakage occurred more frequently following thorascopic surgery, whereas pneumonia was more common following open surgery when comparing complications from both procedures.

Our study was limited by the fact that it only compared data from two national surveys on EA/TEF without statistical validation. Additionally, long-term follow-up data were missing. The divergent assessment criteria used in our surveys prevented a direct comparison of results across the two groups. The follow-up durations in these surveys were insufficient to fully assess postoperative outcomes in EA/TEF patients, representing another limitation. For more accurate outcomes analyses and interpretations in the future, addressing this issue is essential. Establishing assessment criteria at a KAPS meeting beforehand is essential for precise data collection. Despite this, the national surveys offer a current overview, along with extensive details on treatments and outcomes for Korean pediatric patients with EA/TEF. As such, they are expected to be a valuable resource and reference for pediatric surgeons seeking to enhance their understanding of EA/TEF and its treatment options. Statistical analysis and comparison of open versus thorascopic surgery as treatment options for EA/TEF in the forthcoming third national survey would be valuable.

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