

Our Experience with Esophageal Atresia and Tracheoesophageal Fistula

Özefagus Atrezisi ve Trakeoözefageal Fistül Deneyimlerimiz

¹Ali Erdal Karakaya, ¹Ahmet Gokhan Guler, ²Ahmet Burak Dogan, ³Serkan Arslan

¹Department of Pediatric Surgery,
Kahramanmaraş Sutcu Imam University
Faculty of Medicine, Kahramanmaraş,
Turkey

²Department of Pediatric Surgery, Erciyes
University Faculty of Medicine, Kayseri,
Turkey

³Department of Pediatric Surgery, Dicle
University Faculty of Medicine, Diyarbakir,
Turkey

Abstract

To evaluate the treatment results of patients who were operated with the diagnosis of esophageal atresia and tracheoesophageal fistula (EA and TEF) in our clinic. The results of patients who were operated with the diagnosis of esophageal atresia and tracheoesophageal fistula between April 2017 and November 2019 were retrospectively evaluated. The patients were evaluated in terms of gestational age, birth weight, gender, atresia type, surgical approach, and duration of treatment. Postoperative mechanical ventilator follow-up was examined in terms of the transition time to nasogastric tube and oral feeding, and complications. A total of 24 patients, 20 (83%) boys and 4 (17%) girls, were included in the study. The mean age at surgery was 3.9 days. The gestational age of the patients was 35 weeks and the mean birth weight was 2391 grams. Distal tracheoesophageal fistula + proximal atresia were detected in 22 (92%) patients. Congenital heart disease was detected in 14 (58%) patients. The follow-up time in the ventilator was 27 hours, and the average oral feeding time was 11 days. Three (12.5%) patients died. Hospital stay was 16 days. During their follow-up, anastomotic stenosis was observed in 5 (21%) patients, and anastomotic leakage was observed in one (4%) patient. EA and TEF are anomalies that can be highly cured with surgical treatment. The follow-up of patients with EA by the neonatal team is important for the follow-up and treatment of additional anomalies. Therefore, these patients should be followed up with a multidisciplinary approach. Families should be informed about the frequent postoperative anastomotic stenosis.

Keywords: Esophageal atresia; tracheoesophageal fistula; treatment; complications

Özet

Kliniğimizde özefagus atrezisi ve trakeaözefageal fistül (ÖA ve TÖF) tanısı ile ameliyat edilen hastaların tedavi sonuçlarını değerlendirmek. Nisan 2017 - Kasım 2019 tarihleri arasında özefagus atrezisi ve trakeaözefageal fistül tanısıyla ameliyat edilen hastaların sonuçları retrospektif olarak değerlendirildi. Hastaların gestasyonel yaş, doğum ağırlığı, cinsiyet, atrezi tipi, cerrahi yaklaşım, tedavi süresi açısından değerlendirildi. Postoperatif mekanik ventilatör takibi, nazogastrik tüp ve oral beslenmeye geçiş süresi ve komplikasyonlar açısından incelendi. Çalışmaya 20 (% 83) erkek ve 4 (% 17) kız toplam 24 hasta dahil edildi. Ortalama ameliyat yaşı 3,9 gün idi. Hastaların gestasyonel yaşı 35 hafta ve ortalama doğum ağırlığı 2391 gramdı. Distal trakeaözefageal fistül + proksimal atrezi 22 (% 92) hastada tespit edildi. Ondört (% 58) hastada konjenital kalp hastalığı tespit edildi. Ventilatörde takip süresi 27 saat, ortalama oral beslenme süresi 11 gündü. Üç (%12.5) hasta öldü. Hastanede kalış süresi 16 gündü. Takiplerinde 5 (% 21) hastada anastomoz darlığı, bir (% 4) hastada anastomoz kaçağı görüldü. ÖA ve TÖF cerrahi tedavi ile yüksek oranda iyileştirilebilen anomalilerdir. ÖA hastalarının yenidoğan ekibi tarafından izlenmesi, ek anomalilerin takibi ve tedavisi için önemlidir. Bu nedenle bu hastalar multidisipliner bir yaklaşımla takip edilmelidir. Ameliyat sonrası sık karşılaşılan anastomoz darlığı açısından aileler bilgilendirilmelidir.

Anahtar Kelimeler: Özefagus atrezisi; trakeaözefageal fistül; tedavi ; komplikasyonlar

Correspondence:

Ali Erdal KARAKAYA
Department of Pediatric Surgery,
Kahramanmaraş Sutcu Imam
University Faculty of Medicine,
Kahramanmaraş, Turkey
e-mail: dralierdal@gmail.com

Received 30.12.2020 Accepted 15.03.2021 Online published 16.03.2021

Cite this article as:

Karakaya AE, Guler AG, Dogan AB, Arslan S, Our Experience with Esophageal Atresia and Tracheoesophageal Fistula, Osmangazi Journal of Medicine, 2021; 43(4):358- 363 Doi: 10.20515/otd.848843358

1. Introduction

Esophageal atresia (EA) and tracheoesophageal fistula (TEF) are serious, life-threatening diseases in pediatric surgery. EA is seen in 1 / 3000-5000 live births. As a result of developments in neonatal intensive care unit (NICU) and surgical advances in recent years, the chance of survival of these patients has increased. However, there has been a parallel increase in complications of these patients. Early complications that can be seen after surgery include anastomotic leak, anastomotic stenosis and recurrent TEF (1,2). Gastroesophageal reflux (GER) and esophageal dysmotility are frequent postoperative late complications. These complications affect the nutritional intake, growth and development of the infant. As EA often accompanies multiple anomalies, a multidisciplinary approach is important. Additional problems, such as additional anomalies, low birthweight and prematurity of the patient affect morbidity and mortality (3-5)

The aim of this study was to evaluate the treatment results of patients who underwent surgery for a diagnosis of EA and TEF in our

clinic and to present our clinical experience on this subject.

2. Material and Methods

The study was approved by the Local Ethics Committee (Date, no: 21.12.2020/03). A retrospective review was made of the outcomes of patients who were operated on because of EA and TEF between April 2017 and November 2019. The diagnosis of EA was made by nasogastric tube or pouch radiography (Figure 1). All patients were hospitalized in the NICU before and after surgery and were followed up by neonatologist. If the patient had no pathology requiring emergency surgery, surgery was performed under semi-elective conditions. If the patient was not stable, stabilization was achieved first. The patients were evaluated in terms of gestational age at birth, birthweight, gender, atresia type, surgical approach, duration of postoperative mechanical ventilator follow-up, complications.



Figure 1. Pouch radiography

Statistical analysis

Data obtained in the study were analyzed statistically using SPSS vn.17.0 software (IBM Statistics for Windows version 17, IBM Corporation, Armonk, NY, USA). Conformity of continuous data to normal distribution was tested with the Kolmogorov - Smirnov test. Categorical data were stated as n (number) and percentage (%), and quantitative data as mean \pm standard deviation (SD).

3. Results

Evaluation was made of a total of 24 patients, comprising 20 (83%) males and 4 (17%) females, average age of surgery 3.9 days. The average gestational age at birth was 35 weeks, and average birthweight was 2391 grams. Distal TEF + proximal atresia was determined in 22 (92%), isolated esophageal atresia in one (4%) and isolated TEF in one (4%) patients. Congenital anomalies were determined in 18 of 24 (75%) patients. In some patients, it was detected more than one congenital anomaly; congenital heart disease was determined in 14

(58%) patients, urinary system anomaly in seven (29%), total situs inversus in one (4%), duodenal atresia in one (4%), and small for gestational age in one (4%) (Figure2). Thoracotomy was performed in 23 (96%) patients. The primary esophageal anastomosis was performed in one of these patients who isolated esophageal atresia six months after gastrostomy. Right thoracotomy was performed in 16 (70%) of 23 patients who underwent thoracotomy, and muscle-sparing right thoracotomy was performed in seven (30%). The patient with isolated tracheoesophageal fistula was repaired by approaching from the neck. The mean follow-up on the ventilator was 27 hours, mean nasogastric feeding time was 1.5 days and the mean oral feeding time was 11 days. Mortality developed in two patients due to heart disease and in one patient due to prematurity. The mean hospital stay of the patients was 16 days. There were no complications in 18 (75%) patients during follow-up. Anastomotic stenosis was observed in five (21%), and anastomotic leakage was observed in one (4%) patient (Table 1).



Figure 2. Esophagogram: stenosis in the patient with situs inversus

Table 1. Demographic data and results of patients

Gender (M/F)		20 (83%) / 4 (17%)
Surgery age (day)		3.95±6.85
Gestational age		35±3.1
Birth weight (gr)		2391±687
Atresia type	Distal TEF + proximal atresia	22 (92%)
	Isolated TEF	1 (4%)
	Isolated Atresia	1 (4%)
Additional Anomalies <i>Totally 18/24 (75%)</i>	Congenital heart diseases	14 (58%)
	Urinary System Anomalies	7 (29%)
	Gastrointestinal anomalies	1(4%)
	Small for gestational age	1(4%)
Surgical Method	Right thoracotomy	16 (67%)
	Right thoracotomy + muscle sparing	7 (29%)
	Gastrostomy	1(4%)
Nasogastric feeding time (day)		1.5(1-2)
Oral feeding time (day)		11.36±10.50
Ventilator follow-up time (hour)		27.13±25.89
Discharge time (day)		16
Mortality		3 (13%)
Complication	No	18 (75%)
	Stenosis	5 (21%)
	Anastomosis leak	1 (4%)

M:Male, F:Female

4. Discussion

Preoperative risk assessment is important for patients. Developments in anesthesia procedures, intensive care conditions, and nutritional support for these patients have increased the survival of these patients. Low birthweight, prematurity, and concomitant cardiac anomalies are the most important risk factors. Therefore, these risks must be managed well (4). In this study, all patients were followed up in the NICU. Additional anomalies were investigated and pathologies requiring early intervention were investigated. After the operation, the newborn was followed up in the intensive care unit (5,6). As there have been rapid developments in neonatal care, the follow-up of these patients before and after surgery by neonatology specialists can be considered to contribute positively to the success of their treatment. Medical follow-up of the patients in this study was conducted by neonatologist.

The incidence of additional anomalies varies between 20% and 50%, and anomalies of the cardiovascular system are the most common

(7,8). In the current study, the most common pathologies were found to be of the cardiovascular system, similar to reports in literature.

The most common clinical conditions that may be risk factors before surgery are low birthweight and prematurity (9,10). In the current study, mortality developed due to prematurity in one (4%) patient.

In EA cases, thoracotomy or thoracoscopic surgery can be performed as a surgical method (11). Although the thoracoscopic approach is recommended in the current literature, it has the disadvantages of technical difficulty, the need for much experience, and the difficulty in obtaining the necessary surgical equipment. In the more recent cases in this study, muscle-sparing procedures were performed in patients who underwent thoracotomy. This provides advantages such as faster recovery and a shorter surgical procedure.

The tracheal part of the tracheal fistula can be closed with one by one or with transfix suture (7). The tracheal opening of the fistula was closed with a single suture transfix suture in all patients, and no recurrent fistula was observed in any patient. This method was considered to be useful in fistula ligation.

Tracheomalacia can be seen in some EA cases, and has been reported in literature at a rate of 10-20% (12). In the current study, tracheomalacia was observed in 4 (%16) patients, all of which spontaneously improved over time. Aortopexy was not needed in any patient. Tracheomalacia should be considered and investigated in patients with complaints such as bruising and coughing while feeding.

GIS anomalies accompanying EA should be diagnosed before surgery (4). Duodenal atresia was detected in one patient in our clinic and was operated on simultaneously, with no problems. In the repair of EA, our first choice is to perform primary or delayed primary repair according to anatomic type and distance between the ends (13). In the current study, primary anastomosis was performed in 23 patients and delayed anastomosis in one.

Early oral nutrition in newborns significantly reduces morbidity (14). Feeding was started for all the current study patients from the trans anastomotic tube within the first 24 hours after surgery. This can be considered to have contributed to the early recovery of the patients. In addition, efforts were made not to put intubated patients to sleep as far as possible. Therefore, early extubation was possible in patients with suitable clinical conditions.

Complications such as anastomotic stricture, anastomotic leakage, GER, and esophageal dysmotility are the most common complications in patients with EA. The most common complication is anastomotic stenosis, which is seen at the rate of 6-64% (14). In the current study, anastomotic stenosis was observed at a rate of 20%, which is relatively low compared to the literature. The reason for this is that the GAP in between is close and the proximal and distal esophagus are mobilized as much as possible. A tight anastomosis increases complications such as anastomotic stenosis or leakage. Therefore, efforts were made not to have a tense anastomosis to be able to reduce complications in these patients.

One of the early complications of EA is anastomotic leakage, which has been reported in literature at rates of 10%-34% (15,16). In the current study, only one (4%) patient had anastomotic leakage. The reason for this very low rate compared to the literature was that the esophageal gap was narrowed.

5. Conclusion

EA and TEF are anomalies that can be highly cured with surgical treatment. Monitoring of EA patients by the neonatal team is important for the follow-up and treatment of additional anomalies. Therefore, these patients should be followed up with a multidisciplinary approach. Families should be informed about the frequent postoperative anastomotic stenosis.

REFERENCES

1. Taşkınlar H, Kılılı İ, Çelik Y, Avlan D, Özefagus atrezisi ve trakeaözefageal fistüllü hastalardaki deneyimlerimiz. *Çocuk Cerrahi Dergisi* 2012; 26: 32-6.
2. Celayir S, İlçe Z, Tekand GT, Emir H, et al. Özefagus Atrezili Olgularla ilgili 22 yıllık deneyim (1978-2000). *Cerrahpaşa Tıp Dergisi* 2002; 33: 86.
3. Dağlı T. Özefagus atrezisi ve trakeaözefageal fistül. *Türkiye Klinikleri* 2008;2:19
4. Başaklar AC, Türkyılmaz Z. Konjenital özefagus atrezisi ve trakeaözefageal fistül, In: Başaklar AC editor, *Bebek ve Çocukların Cerrahi ve Ürolojik Hastalıkları. Palme Yayıncılık, Ankara;* p: 311-352.
5. Okumuş M. Özefagus atrezisi: Tek Cerrah, Tek Merkez Deneyimi ve Sonuçları. *ACU sağlık Bilimleri Derg* 2020; 11: 264-68.
6. 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.
7. Spitz L: Esophageal atresia and tracheoesophageal malformations, in Ashcraft KW, Holcomb GW, Murphy JP (eds). *Pediatric Surgery.* Philadelphia, *WB saunders* 2005, p 352-70.
 8. Solak H., Göğüs cerrahisi. Konya: *Atlas Kitabevi.* 1993
 9. Vukadin M, Savic D, Malikovic A, Jovanovic D, Milickovic M, Bosnic S, Vlahovic A. Analysis of Prognostic Factors and Mortality in Children with Esophageal Atresia. *Indian J Pediatr.* 2015; 82: 586-90.
 10. 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
 11. Lee S, Lee SK, Seo JM. Thoracoscopic repair of esophageal atresia with tracheoesophageal fistula: overcoming the learning curve. *J Pediatr Surg.* 2014 ;49:1570-2.
 12. Spitz L. Esophageal atresia. Lessons I have learned in a 40-year experience. *J Pediatr Surg.* 2006;41:1635-40.
 13. Spitz L. Oesophageal atresia. *Orphanet J Rare Dis.* 2007; 2:24-25.
 14. Terek D, Yalaz M. Yenidoğan Bebeğin Beslenmesinde Temel Prensipler. *Klinik Tıp Pediatri Dergisi* 2016; 8: 1-13.
 15. Chittmitrapap S, Spitz L, Kiely EM, et al. Anastomotic leakage following surgery for esophageal atresia. *J Pediatr Surg.* 1992 ;27:29-32.
 16. Reusens H, Matthyssens L, Vercauteren C, et al. Multicentre survey on the current surgical management of oesophageal atresia in Belgium and Luxembourg. *J Pediatr Surg.* 2017;52:239-246.