ORIGINAL ARTICLE

A different approach to leakage of esophageal atresia in children

Erol Basuguy, M.D., Mehmet Hanifi Okur, M.D., Serkan Arslan, M.D., Hikmet Zeytun, M.D., Bahattin Aydoğdu, M.D.

Department of Pediatric Surgery, Dicle University Faculty of Medicine, Diyarbakır-*Turkey*

ABSTRACT

BACKGROUND: In this study, we aimed to present the results of patients treated for esophageal leakage with a different conservative approach.

METHODS: Ninety-eight patients with esophageal atresia and tracheoesophageal fistula (EA) who underwent surgery in our clinic between February 2013 and January 2018 were retrospectively reviewed in this study. Patients' anastomosis leakage, gestational week, gender, body weight, referral date, recovery time and stenosis were recorded. After leakage detection, the nasogastric catheter was fluoroscopically converted into a nasojejunal catheter using a guidewire and feeding continued.

RESULTS: Anastomotic leakage developed in 18 (18.3%) patients. The average gestational age at birth was 35.4 weeks; the patients included ten girls and eight boys of average weight 2.41 kg; the average referral period was 2.1 days after birth and the average time of surgery was 2.4 days after birth. The average recovery time was 21.1 days (range: 8–60 days). Eight patients developed stenosis that recovered with dilatation.

CONCLUSION: Our findings suggest that our conservative treatment approach, which uses a nasojejunal catheter, is an effective method that would reduce complications, enable earlier feeding, and reduce the cost compared to other treatment approaches.

Keywords: Children; esophagus; leakage; nasojejunal catheter.

INTRODUCTION

Newborn esophageal atresia (EA) and trachea-esophageal fistulae (TEF) are relatively common congenital abnormalities with incidence rates of one in 2,500 and one in 4,500 live births.[1,2] Improvements in neonatal intensive care, anesthesia, and surgical techniques have considerably improved the survival rates of the patients with EA.[3] However, as the number of operations increases, the number of postoperative complications inevitably increases; therefore, the complications remain a concern.[4,5] One of the most common and severe complications following EA/TEF surgery is esophageal leakage; the incidence is approximately 15–17%.[6] Despite recent developments in pediatric surgery, some patients require re-operation, which remains a major surgical concern because the associated morbidity and mortality rates are relatively high. In this study, we aimed to present the results of patients treated for esophageal leakage with a different conservative approach.

MATERIALS AND METHODS

Ninety-eight patients with esophageal atresia and tracheoesophageal fistulae who underwent surgery at our clinic between February 2013 and January 2018 were retrospectively reviewed in this study. Patients with isolated EA were excluded from this study. All cases were graded using the Gross classification and surveyed using the SPITZ system. After preoperative stabilization, all patients were reviewed concerning genitourinary, cardiovascular, anorectal, and spinal abnormalities. Gestational week, gender, body weight, and referral date were recorded. Gap length was measured with a ruler

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Address for correspondence: Erol Basuguy, M.D.
Dicle Üniversitesi Tıp Fakültesi, Çocuk Cerrahisi Anabilim Dalı, Diyarbakır, Turkey
Tel: +90 412 - 248 80 01 / 4718 E-mail: erbas.80@hotmail.com

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during an operation before dissecting the fistula (short gaps: 72 patients, long gaps: 26 patients). We defined as a long gap length of ≥3 cm (the distance between the two ends of the esophagus).[7] Two groups were compared concerning leakage, stricture rates and leakage recovery time. Anastomotic leakage developed in 18 (18.3%) patients. The need for mechanical ventilation of these patients and the recovery times were recorded. All patients underwent thoracotomy using the right intercostal space (ICS) 4–5 while lying on the left. Continuously feeding was started 1–2 mL per hour of breast milk or formula using a nasogastric catheter on postoperative day 2, and the quantity of milk was gradually increased daily. Esophagography was performed on all patients who exhibited no thoracic tube discharge on postoperative day five and patients without leakage were started oral feeding. Patients were discharged with a recommendation for polyclinic follow-up two weeks later.

Patients who exhibited formula or saliva discharge using the thoracic tube underwent esophagography in a dedicated room at our clinic. After leakage detection (Fig. 1a), the nasogastric catheter was fluoroscopically converted into a nasojejunal catheter using a guidewire and feeding continued (Fig. 1b). The flow rate and nature of thoracic tube discharge were recorded daily. Any lung problems were noted. Secondary tubes were placed in five patients who developed pneumothorax that could not be controlled using a single tube. All radiological examinations were carried out in our clinic and/or at the bedside. Patients were monitored by performing complete blood counts, biochemical analyses, blood gas evaluations, culture antibiograms, and we obtained posterior-anterior lung X-rays. The antibiograms were used to guide the antibiotherapy of patients who developed leakage and to plan new therapies.

Patients who developed leakage were discharged with a recommendation for polyclinic follow-up two weeks after full oral feeding commenced if lung problems were absent. Follow-up esophageal scans were carried out for patients with complaints and patients developing stenosis underwent balloon dilatation under general anesthesia. Our treatment algorithm for the management of anastomotic leakage is shown in Figure 2.

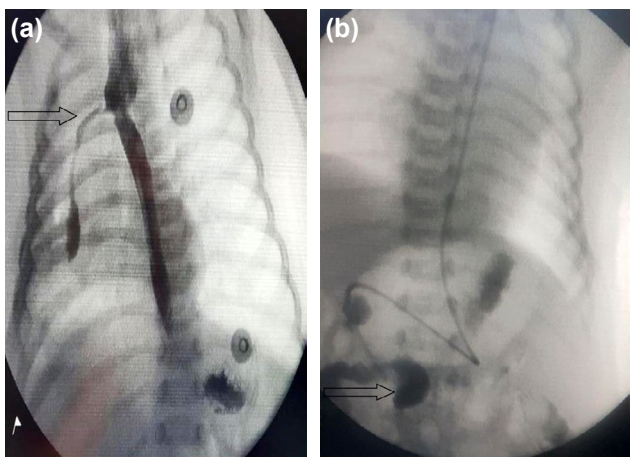

Figure 1. (a) Anastomotic leakage. **(a)** The Nasojejunal catheter.

This study was approved by the Committee on Ethics in Non-interventional Clinical Studies of Dicle University, Faculty of Medicine (June 6, 2018; decision no. 56).

Statistical Analysis

All statistical analyses were performed using SPSS for Windows software (ver. 15.0; SPSS Inc., Chicago, IL, USA). The chi-squared test was used to compare categorical data. The Kolmogorov–Smirnov test was employed to explore if numerical data were normally distributed; such data were compared using the t-test, whereas data with non-normal distributions were compared using the Mann–Whitney U test. A p-value <0.05 was considered to reflect statistical significance.

RESULTS

On gross EA classification, 17 patients were of type C and 1 of type D (with both a distal and proximal fistula). On SPITZ classification, 83.3% (n=15) of 18 patients who developed leakage were of Group 1, 11% (n=2) of Group 2, and 5.5% (n=1) of Group 3.

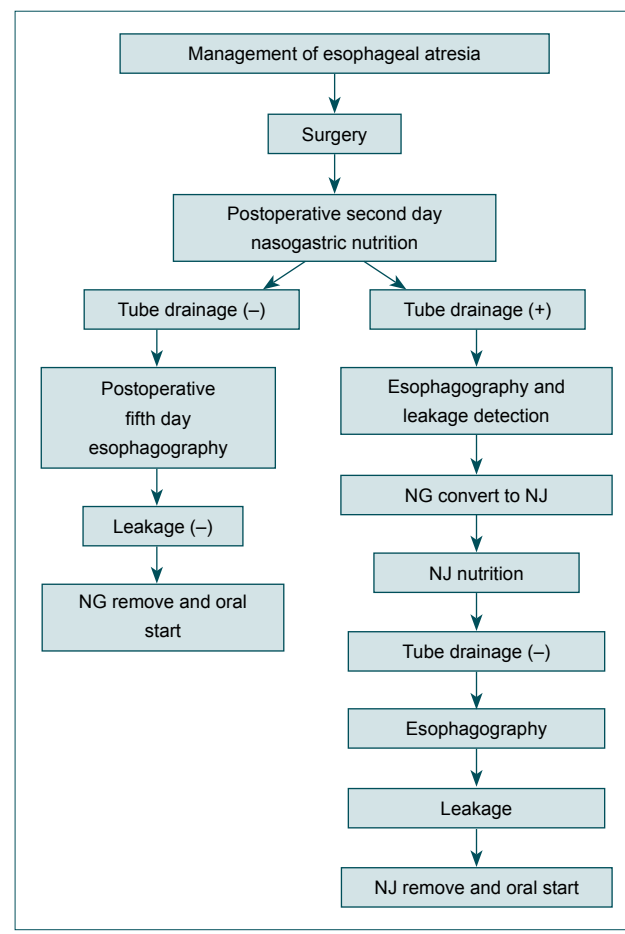

Figure 2. Our treatment algorithm.

Table 1. Demographic data of the patients who developed leakage

	Short gap	Long gap
	n (%)	n (%)
Patients	10 (14)	8 (31)
Gender		
Male	4	4
Female	6	4
Gestasyonel age	34.94	36.13
Weight	2.29	2.56
Additional anomaly		
Cardiac anomaly	5 (27.7)	2 (11)
Urogenital system	1	1
Gastrointestinal system	1	1
Mechanical ventilation	3	2
Leakage recovery time (day)	16.05	27.66
Infection	2 (11)	3 (16.6)
Stricture	3 (16.6)	5 (27.7)

The average gestational age at birth was 35.4 weeks; the patients included 10 girls and eight boys of average weight 2.41 kg; the average referral period was 2.1 days after birth, and the average time of surgery was 2.4 days after birth. Of the patients who developed leakage, eight (31%) patients had long gaps, and 10 (14%) patients had short gaps. The average recovery time was 21.1 days (range: 8–60 days).

The most common additional problem was cardiac abnormalities in seven (38.8%) patients, of whom two patients exhibited genitourinary, two patients gastrointestinal, two patients limb, and one patient central nervous system abnormalities. One patient had VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities).

In total, five patients who developed leakage were intubated due to infection and lung problems. Signs of infection appeared in five patients during treatment; the treatment plans were thus changed. Eight patients developed stenosis, including five with long and three with short gaps; all underwent balloon dilatation. Multiple dilatations were performed to three patients and single balloon dilatations to five patients. Leakage developed in one patient after balloon dilatation and was corrected by conservative treatment. No problem was observed during the follow-up of patients who underwent dilatation. TEF relapse developed in one patient; surgical repair was performed. Demographic data, the details of the abnormalities, and the follow-up status of all patients are listed in Table 1.

DISCUSSION

Anastomotic leakage is one of the most common severe complications after esophageal atresia (EA) repair. Despite improvements in the surgical correction of EA, postoperative leakage continues to be a significant problem. Leakage is affected by many factors, including esophageal injury, excessive mobilization of the distal end of the esophagus, associated ischemia,[8] anastomotic tension,[9] poor suturing technique, the use of inappropriate sutures,[10,11] and a long gap length and sepsis.[8–11] Of these, the gap length is of particular concern, being an independent risk factor for leakage, a longer gap length has been suggested to correlate with increased leakage. Reported gap lengths vary; however, a gap length of ≥3 cm (the distance between the two ends of the esophagus) is accepted as being long.[7]

A previous study suggested that the leakage rate increased with gap length; the rate was 30% (15/50) in the cited work.[12] In contrast, Thakkar et al.[13] suggested that the gap length was not associated with the anastomotic leakage or stricture rate. However, we found that leakage, stricture rate, and leakage recovery time were significantly higher in patients with long gaps ($p<0.05$).

Although different treatment approaches have been proposed, the optimal monitoring and treatment of leakage remain controversial. Some studies have recommended conservative treatment; other studies suggested that surgery is essential. However, the optimal surgical technique is still unclear.

Some authors recommend early thoracotomy and re-anastomosis to optimize outcomes, whereas other authors favored gastric pull-up or colonic or jejunal transposition.[14–16] Koivusalo et al.[17] re-operated on 17 out of the 20 patients who developed leakages; anastomotic strictures developed in four patients, and one patient died. Chittmittrapap et al.[10] reported that esophageal leakage developed in 34 (17%) patients and six out of seven undergoing re-operation lost the native esophagus due to major disruptions. Early thoracotomy was recommended for such patients. Bawa et al.[18] monitored patients with esophageal leakage for an average of 12 days and then performed gastrostomy and jejunostomy (because of nutritional concerns) to treat patients in whom leakage persisted. Three of the patients developed jejunostomy tube-related complications and six patients died.

Reoperation is considered dangerous due to continuous exposure of the tissue to gastric fluid and secretions in the case of postoperative leakage and may indicate that thoracotomy and re-anastomosis may be inappropriate. Other concerns include possible loss of the native esophagus, poor motility, reflux, and leakage.[6,15,19]

Conservative treatments include chest tube placement, total parenteral nutrition (TPN), and wide-spectrum antibiotics.[20] Huang et al.[21] reported that 9% (n=3) of patients who underwent operations developed leakage; all recovered with conservative treatment.

Vaghela et al.[22] reported that the use of glycopyrrolate, a secretion-reducing agent, was associated with more rapid leakage resolution, reduced need for mechanical ventilation, earlier enteral nutrition, and better preservation of the natural esophagus compared to the other group. They also suggested that enteral nutrition using slow infusion reduced gastroesophageal reflux (GER). However, secondary operations were required for two out of 21 patients in this group and the side-effects of this drug included constipation, urinary retention, dry mouth, and moniliasis.

TPN may cause cholestasis, and a central venous catheter is required if TPN is long-term. Both catheter-associated complications and cardiovascular events may develop, increasing morbidity and mortality.[23–26] A previous study considered that enteral nutrition was more physiological, easier, and cheaper than TPN, without any need for a central venous catheter. This type of nutrition requires fewer laboratory tests and is associated with fewer complications.[27]

Our treatment protocol was conservative in all patients, and this approach was noted in our previous clinical study.[28] We delivered enteral nutrition using a nasojejunal catheter after leakage detection, chest tube placement, and commencement of antibiotics. A nasojejunal catheter can be easily placed in newborns with the aid of scope; there is no need for anesthesia, and earlier enteral feeding is possible.

Our approach is physiological and patient-friendly; all required calories are delivered. Enteral nutrition did not trigger GER, although the quantity of food was increased daily. Therefore, TPN was not used. We thus prevented possible complications, such as cholestasis, cardiovascular events, and infection, arising from the use of a central catheter. Furthermore, the cost was able to be reduced since no TPN or central venous catheter was used. We believe that this approach, without any additional drugs or secondary surgery, effectively reduces complications and enables earlier feeding. Furthermore, the native esophagus is preserved.

Earlier studies reported mortality rates of approximately 30%; half of all mortality was caused by sepsis associated with the leakage of long gaps.[12] We performed preoperative risk classification using the Spitz system. Two patients (11.8%) died: No patient died in Group 1, one patient died in Group 2, and one patient died in Group 3. No patient died because of leakage per se or complications that arise from leakage.

Conclusion

Surgery performed to esophageal leakage may trigger severe complications, such as a difficult anastomosis, leakage, poor motility, and reflux, due to tissue fragility, the preferred approach remains conservative treatment. TPN, central catheter and the use of glycopyrrolate are possible risk factors in patients in whom conservative treatment is performed concerning mortality and morbidity. We believe that our conservative treatment approach, which uses a nasojejunal catheter, is an effective method that would reduce these complications, enable earlier feeding, and reduce the cost.

Acknowledgments

The authors declare that there is no conflict of interest regarding the publication of this article.

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ORİJİNAL ÇALIŞMA - *ÖZET*

Çocuklarda özofagus atrezisi kaçaklarına farklı bir yaklaşım

Dr. Erol Basuguy, Dr. Mehmet Hanifi Okur, Dr. Serkan Arslan, Dr. Hikmet Zeytun, Dr. Bahattin Aydoğdu

Dicle Üniversitesi Tıp Fakültesi, Çocuk Cerrahi Anabilim Dalı, Diyarbakır

AMAÇ: Farklı bir konservatif yaklaşımla özofagus kaçağı nedeniyle tedavi edilen hastaların sonuçlarını sunmayı amaçladık.
GEREÇ VE YÖNTEM: Kliniğimizde Şubat 2013–Ocak 2018 tarihleri arasında trakeoözofageal fistüllü özofagus atrezisi (EA) nedeniyle ameliyat edilen olan 98 hasta geriye dönük olarak incelendi. Anastomoz kaçağı olan hastalarda gebelik haftası, cinsiyet, vücut ağırlığı, başvuru günü, iyileşme süresi ve stenoz kaydedildi. Kaçak tespitinden sonra, nazogastrik kateter floroskopik olarak bir kılavuz tel kullanılarak nazojejunal katetere dönüştürüldü ve beslenme devam etti.
BULGULAR: Anastomoz kaçağı 18 (%18.3) hastada gelişti. Ortalama gebelik yaşı yaşı 35.4 haftaydı; hastaların onu kız ve sekizi erkekti. Ortalama ağırlık 2.41 kg idi. Başvuru günü doğumdan sonra ortalama 2.1 gün ve ortalama ameliyat zamanı doğumdan sonra 2.4 gün idi. Ortalama iyileşme süresi 21.1 gündü (8–60 gün). Sekiz hastada dilatasyon ile iyileşen darlık gelişti.
TARTIŞMA: Nazojejunal kateter kullanılarak yapılan konservatif tedavi yaklaşımımızın diğer tedavi yaklaşımlarına kıyasla komplikasyonları azaltıp, erken beslenmeyi sağlayacak ve maliyeti düşürecek etkili bir yöntem olduğuna inanıyoruz.
Anahtar sözcükler: Çocuklar; kaçak; nazojejunal kateter; özofagus.

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