

## Sealing of esophageal perforation with a fully covered biliary stent in a pediatric patient

Mustafa Okumuş

Department of Pediatric Surgery, Yeni Yüzyıl University, Faculty of Medicine, Gaziosmanpaşa Hospital, İstanbul, Turkey

ORCID ID of the author(s)

MO: 0000-0001-8705-5183

### Abstract

Conservative treatment for esophageal perforations can cause problems related to both nutrition and wound healing in pediatric patients due to its long duration. This case report presents a 14-month-old female patient who underwent endoscopic balloon dilatation for esophageal stricture. The patient had been operated on for esophageal atresia in the neonatal period. Eight hours after discharge, the patient was hospitalized again due to esophageal perforation. Although conservative treatment lasting three weeks was the preferred method, it was unsuccessful. Therefore, a fully covered biliary stent was used instead of an esophageal stent, as the appropriate size stent could not be found. The stent sealed the perforation, and the patient started to be fed orally on the third day. The esophageal stent was removed on the 17th day, and no leakage was observed on the esophagogram. Although conservative methods are the first-line treatment for esophageal perforations in children, their long duration and the inability to feed for a long time are significant disadvantages. Fully covered self-expandable esophageal stents may be a reliable alternative for sealing esophageal perforations in pediatric patients, as they are in adults.

**Keywords:** esophageal perforation, conservative treatment, esophageal stent, fully covered self-expandable metallic stents, biliary stents, pediatrics

### Introduction

Iatrogenic esophageal perforation is a rare but serious complication that pediatric surgeons hope to avoid. It is mainly caused by therapeutic procedures performed endoscopically. Esophageal perforation can lead to various consequences, including localized para-esophageal abscess, diffuse mediastinitis, empyema, and even death [1]. As such, early diagnosis and treatment are vital.

Conservative methods are typically the preferred treatments for early-diagnosed and hemodynamically stable patients with esophageal perforations [2]. However, self-expandable esophageal stents and esophageal vacuum treatments are also effective methods [3,4]. If minimally invasive treatments fail or esophageal damage is severe, surgical treatment should always be considered a viable alternative [5].

In this case report, we describe the treatment of a 14-month-old girl who developed esophageal perforation after balloon dilatation. Despite an early diagnosis, conservative treatment lasting three weeks was unsuccessful. However, effective results were achieved within two weeks using a fully covered self-expandable biliary stent as an esophageal stent.

### Corresponding Author

Mustafa Okumuş  
Yeni Yüzyıl University, Faculty of Medicine,  
Department of Pediatric Surgery, Gaziosmanpaşa  
Hospital, Gaziosmanpaşa, İstanbul, Turkey  
E-mail: drmustafaokumus@gmail.com

### Informed Consent

The authors stated that the written consent was obtained from the parents of the patient presented with images in the study.

### Conflict of Interest

No conflict of interest was declared by the authors.

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## Case presentation

A 14-month-old female patient who underwent esophageal atresia surgery during the neonatal period and had an anastomotic stricture underwent esophageal balloon dilatation. The patient was discharged on the same day after feeding without any problems. However, eight hours after discharge, the patient was brought to the emergency department complaining of inability to swallow and vomiting. No pathology was observed on the chest X-ray. The patient was hospitalized, and oral intake was stopped while intravenous fluid and electrolyte therapy were initiated.

The following morning, the patient developed significant respiratory distress, and an esophagogram (Figure 1) revealed esophageal perforation and right pneumothorax. The patient was taken to the operating room, and under general anesthesia, a chest tube, central venous catheter, and nasogastric tube were inserted. We decided not to perform surgical intervention for the primary repair of the esophageal perforation and instead opted for conservative management. The patient was started on broad-spectrum antibiotics and total parenteral nutrition.

On the fourth day, the patient removed the nasogastric tube. We decided not to reinsert it because the tube had been passing directly from the perforation site in the esophagus to the right thoracic cavity. Although the thoracic tube drainage did not decrease during the first week of follow-up, there was no significant change in the patient's general condition. Therefore, we decided to continue with the same treatment.

The patient's general condition was more stable in the second week than in the first. However, esophageal leakage was still present on the esophagogram at the end of the second week. Therefore, we decided to perform an esophagoscopy and insert a nasoduodenal tube. During the rigid esophagoscopy, we observed that the perforation was located just at the upper edge of the stricture and was approximately 5–7 mm in diameter. The relatively small size of the perforation led us to continue with conservative treatment. Although we attempted to feed the patient nasoduodenally, we could not pass the tube to the duodenum. This was a disappointment at the end of the third week.

We decided to place a stent in the esophagus. As no suitable fully covered self-expandable esophageal stent was available, we opted to use a fully covered nitinol biliary stent designed for adults as an esophageal stent. The stent we used was 8 cm in length, with a trunk section diameter of 10 mm and end diameters of 13 mm (Figure 2). A 6-cm stent may have been more suitable, but one was unavailable. To prevent stent migration, we tied it with a string and secured it on the nose side.

The first day after placing the stent was marked by restlessness and retching, leading us to consider removing the stent. However, the patient's symptoms subsided and stabilized on the following day. Chest tube drainage decreased from the first day, and we began feeding the patient orally on the third day after confirming no leakage on an esophagogram. On the seventh day, we removed the chest tube. We removed the esophageal stent on the 17th day, and an esophagogram showed no leakage (Figure 3). The patient continued with oral nutrition and was discharged on the 44th day with full recovery. Despite receiving total parenteral nutrition, the patient experienced significant weight loss of approximately 4–4.5 kg. She had no hemodynamically significant

problems, but we had to transfuse her with red blood cell suspension twice. After six months, she was doing well with no complaints.

Figure 1: Esophageal leakage (black arrow) and right pneumothorax (white arrow: border of the collapsed lung).

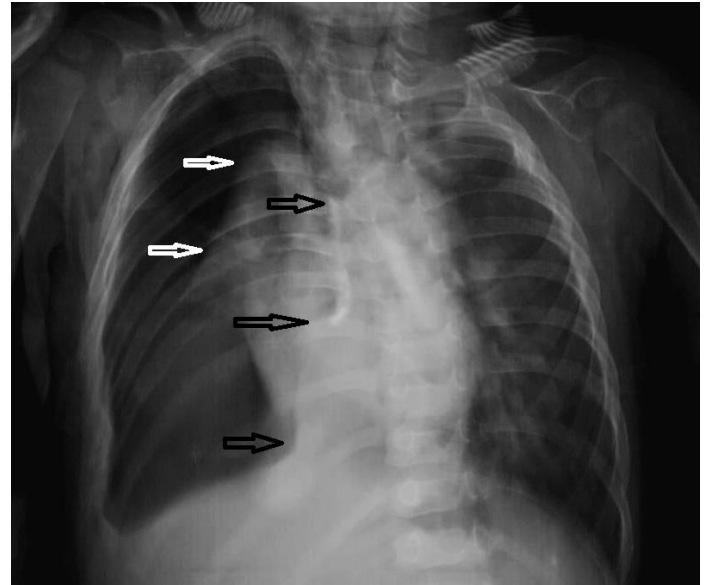


Figure 2: Fully covered self-expanded nitinol biliary stent in the esophagus (black arrow).

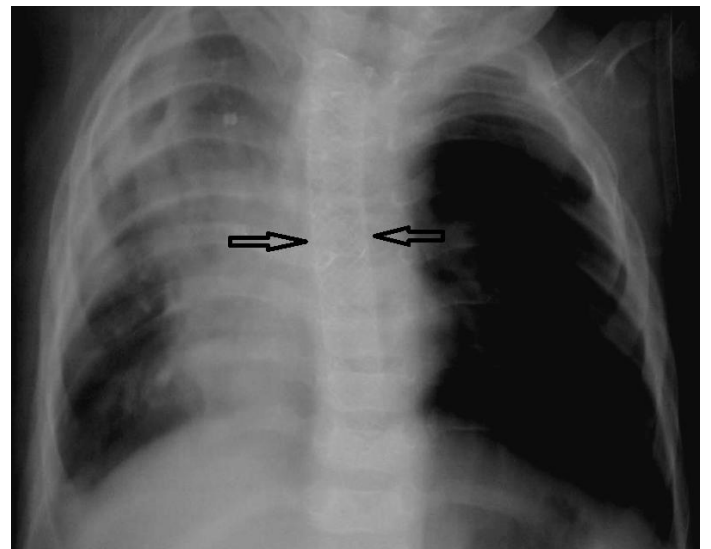
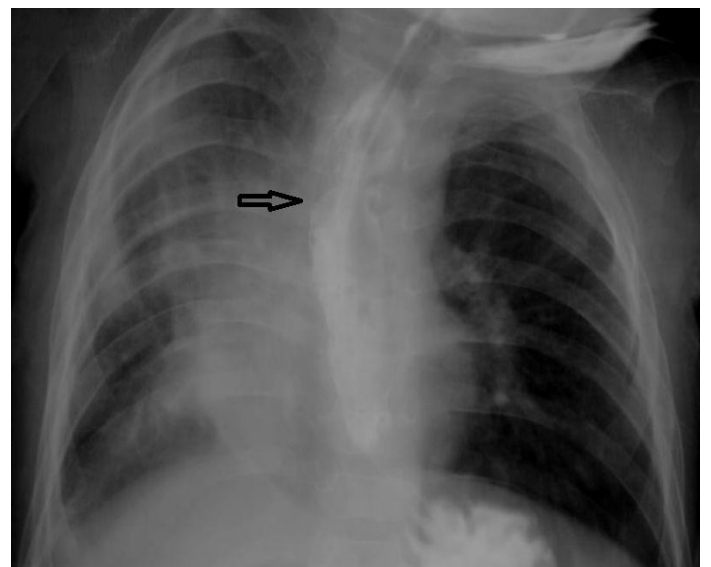


Figure 3: There was no leakage after the stent removal (black arrow: previous leakage site).



## Discussion

Esophageal perforation in children can be caused by blunt injury to the chest or neck, nasogastric tube placement, endotracheal intubation, caustic ingestion, foreign body ingestion, and endoscopic procedures [6]. Iatrogenic causes account for 77% of esophageal perforations, with an incidence rate of approximately 0.6% [7]. Although endoscopy is frequently used for diagnostic and therapeutic purposes in children, esophageal perforation is an extremely rare complication. The most common cause of esophageal perforation is stricture dilatation using the balloon or bougie method, as in the presented case [1].

While the mortality rate for esophageal perforations is around 28%, delayed diagnosis significantly increases morbidity and mortality [1,8]. Therefore, early diagnosis is a crucial factor for successful treatment. Any child who develops symptoms following an endoscopic esophageal procedure should be evaluated for the presence of esophageal perforation [9]. However, not all patients experience respiratory distress, tachycardia, and tachypnea during the early period. In our patient's case, no symptoms were observed during the early period, and she was discharged after normal oral feeding. The chest X-ray taken after the procedure was deemed normal, as was the second chest X-ray taken when she was admitted to the emergency department. In cases of uncertainty, it is essential not to rely solely on a simple chest X-ray, and it should be kept in mind that even an esophagogram may not reveal pathology in 10% of patients [1]. If necessary, tomography and esophagoscopy should also be performed for diagnosis.

The basis of conservative treatment for esophageal perforation includes discontinuing oral intake, administering total parenteral nutrition, initiating broad-spectrum antibiotics, and draining with a chest tube, if necessary. If the patient is hemodynamically stable and without sepsis or esophageal necrosis, aggressive conservative treatment has reportedly resulted in a 100% success rate within a reasonable timeframe [2]. Although the goal of conservative treatment is to preserve the patient's natural esophagus, in life-threatening situations, diversion or esophageal replacement surgeries can be considered [5]. However, what should be done if the patient is hemodynamically stable, and conservative treatment fails to produce results? Should we continue to wait? In the presented case, the patient's clinical condition did not significantly improve during the 3-week conservative treatment period, and nasoduodenal feeding was not feasible. Although nasogastric tube feeding was a possibility, the potential consequences of gastroesophageal reflux were concerning. As a result, the patient developed protein-energy malnutrition and impaired wound healing despite total parenteral nutrition.

Fully covered self-expandable esophageal stents may not be effective in treating stricture dilatations due to their high recurrence rates [10]. However, they have proven effective in treating esophageal perforations [3,11]. Although the recommended treatment time for esophageal stents is 6–8 weeks [12], this period can be shortened for perforation sealing. During this period, the patient can be fed orally with liquid food. While pain and retching are often temporary issues, stent migration may require repositioning, which occurs in 29% of patients [13]. In our case, we did not encounter any issues with stent migration, and we

initiated oral feeding on the third day following esophageal stent placement. It should be noted that while the esophageal stent prevents the passage of saliva to the perforation site and allows for oral feeding, there is a risk of esophageal erosion and pressure on the fragile, traumatized esophageal wall. Therefore, choosing the correct diameter and size for the stent is essential.

Endoscopic vacuum therapy is a promising treatment method for esophageal perforations, with successful results. Studies conducted with children and adults have reported success rates of around 83–88% [4,14]. The median duration of endoscopic vacuum therapy in pediatric patients is 8 days [4], significantly shorter than esophageal stent therapy and conservative therapy.

## Conclusion

Although conservative treatment is still the preferred method for esophageal perforations in hemodynamically stable children, the length of treatment, hospital stay, and nutritional problems are significant disadvantages. Current approaches, such as self-expandable esophageal stents, could be a good alternative in cases where conservative treatment fails to achieve the desired result. In appropriate pediatric cases, fully covered self-expandable biliary stents could be used successfully if necessary.

## References

- Gander JW, Berdon WE, Cowles RA. Iatrogenic esophageal perforation in children. *Pediatr Surg Int*. 2009;25(5):395-401.
- Martinez L, Rivas S, Hernández F, Avila LF, Lassaletta L, Murcia J, et al. Aggressive conservative treatment of esophageal perforations in children. *J Pediatr Surg*. 2003 May;38(5):685-9. doi: 10.1016/j.psu.2003.50183. PMID: 12720170.
- Lange B, Demirakca S, Kähler G, Weiß C, Wessel L, Kubiak R. Experience with Fully Covered Self-Expandable Metal Stents for Esophageal Leakage in Children. *Klin Padiatr*. 2020 Jan;232(1):13-9. English. doi: 10.1055/a-1014-3179. Epub 2019 Oct 16. PMID: 31618788.
- Manfredi MA, Clark SJ, Staffa SJ, Ngo PD, Smithers CJ, Hamilton TE, et al. Endoscopic Esophageal Vacuum Therapy: A Novel Therapy for Esophageal Perforations in Pediatric Patients. *J Pediatr Gastroenterol Nutr*. 2018 Dec;67(6):706-12. doi: 10.1097/MPG.0000000000002073. PMID: 29927863.
- El-Asmar KM, Elghandour MM, Allam AM. Iatrogenic esophageal perforation caused by endoscopic dilatation of caustic stricture: Current management and possibility of esophageal salvage. *J Pediatr Surg*. 2021;56(4):692-6.
- Rentea RM, St Peter SD. Neonatal and pediatric esophageal perforation. *Semin Pediatr Surg*. 2017;26(2):87-94.
- Fernandez FF, Richter A, Freudenberg S, et al. Treatment of endoscopic esophageal perforation. *Surg Endosc*. 1999;13(10):962-6.
- Jones WG 2nd, Ginsberg RJ. Esophageal perforation: a continuing challenge. *Ann Thorac Surg*. 1992;53(3):534-43.
- Panieri E, Millar AJ, Rode H, Brown RA, Cywes S. Iatrogenic esophageal perforation in children: patterns of injury, presentation, management, and outcome. *J Pediatr Surg*. 1996 Jul;31(7):890-5. doi: 10.1016/s0022-3468(96)90404-2. PMID: 8811550.
- Fallon BP, Overman RE, Geiger JD, Jarboe MD, Kunisaki SM. Efficacy and risk profile of self-expandable stents in the management of pediatric esophageal pathology. *J Pediatr Surg*. 2019 Jun;54(6):1233-8. doi: 10.1016/j.jpedsurg.2019.02.025. Epub 2019 Feb 28. PMID: 30890268.
- Rollins MD, Barnhart DC. Treatment of persistent esophageal leaks in children with removable, covered stents. *J Pediatr Surg*. 2012;47(10):1843-7.
- Vermeulen BD, Siersema PD. Esophageal Stenting in Clinical Practice: an Overview. *Curr Treat Options Gastroenterol*. 2018;16(2):260-73.
- Best C, Sudel B, Foker JE, Krosch TC, Dietz C, Khan KM. Esophageal stenting in children: indications, application, effectiveness, and complications. *Gastrointest Endosc*. 2009 Dec;70(6):1248-53. doi: 10.1016/j.gie.2009.07.022. PMID: 19836746.
- Jung CFM, Müller-Dornieden A, Gaedcke J, Kunsch S, Gromski MA, Biggemann L, et al. Impact of Endoscopic Vacuum Therapy with Low Negative Pressure for Esophageal Perforations and Postoperative Anastomotic Esophageal Leaks. *Digestion*. 2021;102(3):469-79. doi: 10.1159/000506101. Epub 2020 Feb 11. PMID: 32045916.