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Successful Late Endoscopic Stent-Grafting in a Patient with Boerhaave Syndrome

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 53-year-old
Final Diagnosis: Spontaneous esophageal rupture
Symptoms: Chest pain • dyspnea • hydropneumothorax • purulent discharge from the umbilicus • vomiting
Medication: —
Clinical Procedure: Endoscopic stent-grafting • enteral feeding • pleural drainage
Specialty: Gastroenterology and Hepatology • Surgery

Objective: Unusual setting of medical care

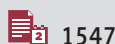
Background: Boerhaave syndrome is a rare esophageal injury associated with a high mortality rate of 14.8%. Immediate diagnosis and treatment have been associated with a better outcome. Surgery remains the mainstay of treatment for those who present early with widespread septic contamination. One of the most difficult dilemmas in the treatment of Boerhaave syndrome is selection of the most appropriate management for late perforations with severe septic complications. In this situation, aggressive surgical approach with esophagectomy and immediate or postponed reconstruction is usually recommended.

Case Report: We report a patient with spontaneous esophageal rupture successfully treated by late endoscopic stent-grafting. The patient was transferred from a rural hospital after initial non-effective conservative treatment. Endoscopic stent-grafting was performed 7 days from the onset of symptoms. A self-expanding plastic stent-graft (Polyflex) used initially very early migrated to the stomach. The plastic stent-graft was then replaced by a fully covered self-expandable metal stent-graft (EndoMAXX), which was wider and equipped with anti-migration struts. Implantation of the EndoMAXX stent-graft resulted in clinical success, with the closure of esophageal rupture confirmed 8 weeks after stent-grafting.

Conclusions: Our case indicates that even late after spontaneous esophageal perforation, less invasive treatment by endoscopic stent-grafting with adequate drainage of septic contamination may be an attractive option for preserving the esophagus in selected patients in stable hemodynamic condition. Our case also supports implantation of wider metallic stent-grafts to seal benign esophageal perforation above the esophago-gastric junction to prevent early migration to the stomach.

Keywords: Boerhaave Syndrome • Esophageal Perforation • Rupture, Spontaneous • Self Expandable Metallic Stents

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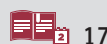
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Background

Boerhaave syndrome is a rare esophageal injury with a diverse presentation and thus is frequently overlooked. A delayed diagnosis results in substantial bacterial and chemical contamination of mediastinal and/or pleural compartment. Severe septic complications contribute to a high mortality rate of 14.8% [1]. Since Dr. Norman Barrett in 1947 introduced surgical repair resulting in a dramatic decrease in mortality, operative therapy traditionally has been recommended to manage patients with Boerhaave syndrome [2-6]. Recently, temporary endoscopic implantation of self-expandable stent-grafts has been proposed for the treatment of esophageal perforation [7-11]. We report a patient with a spontaneous esophageal rupture successfully treated by late endoscopic stent-grafting with plastic and metal self-expandable endoprosthesis.

Case Report

A 53-year-old man presenting sudden chest pain with concomitant dyspnea and vomiting was admitted to a rural hospital. Plain chest X-ray and computed tomography revealed left-sided hydropneumothorax. A chest tube introduced to the left pleural cavity evacuated purulent content with food particles. Suspected spontaneous esophageal rupture was confirmed by endoscopy and a feeding tube was inserted to the duodenum. These interventions improved the patient's condition, and conservative management with chest drainage, enteral feeding, and antibiotic therapy was attempted. The patient was transferred to our institution (a tertiary care and regional center of esophageal surgery) 5 days later due to a persisting large amount of purulent discharge from the pleural cavity (*Acinetobacter baumani* and *Candida albicans* were

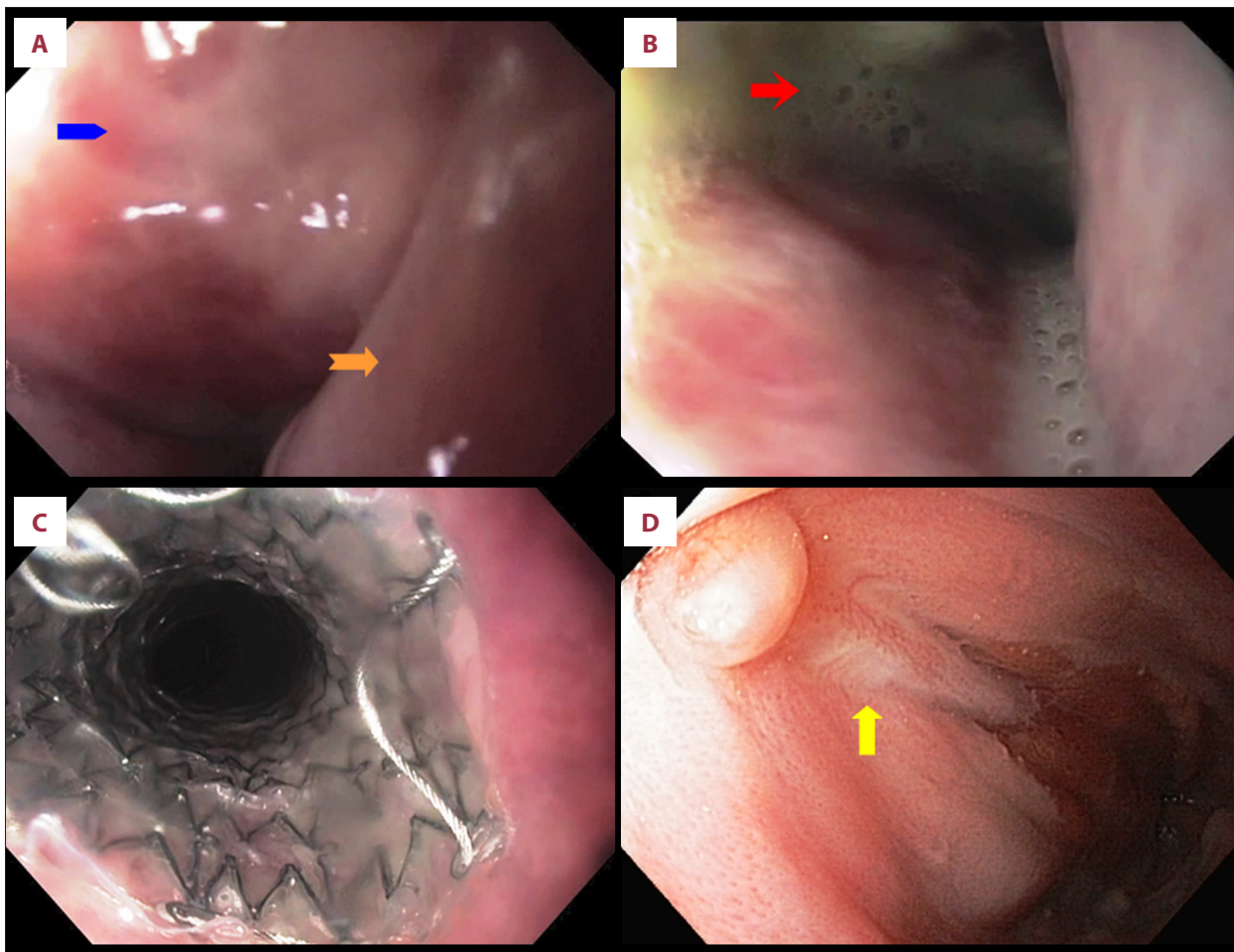


Figure 1. Endoscopic presentation of esophageal perforation in Boerhaave syndrome and its sealing with a self-expandable metallic stent. (A) Demonstrates a complete disruption of the esophageal wall and visible mediastinal tissue (orange sharp-ended arrow indicates the edge of esophageal wall without no detectable pre-existing pathology; blue smooth arrow indicates mediastinal tissue). (B) Demonstrates connection to the left pleural cavity with purulent discharge (red sharp arrow). (C) Demonstrates the EndoMAXX stent after implantation. (D) Demonstrates scar (yellow regular arrow) sealing of the site after esophageal perforation 8 weeks after stenting.

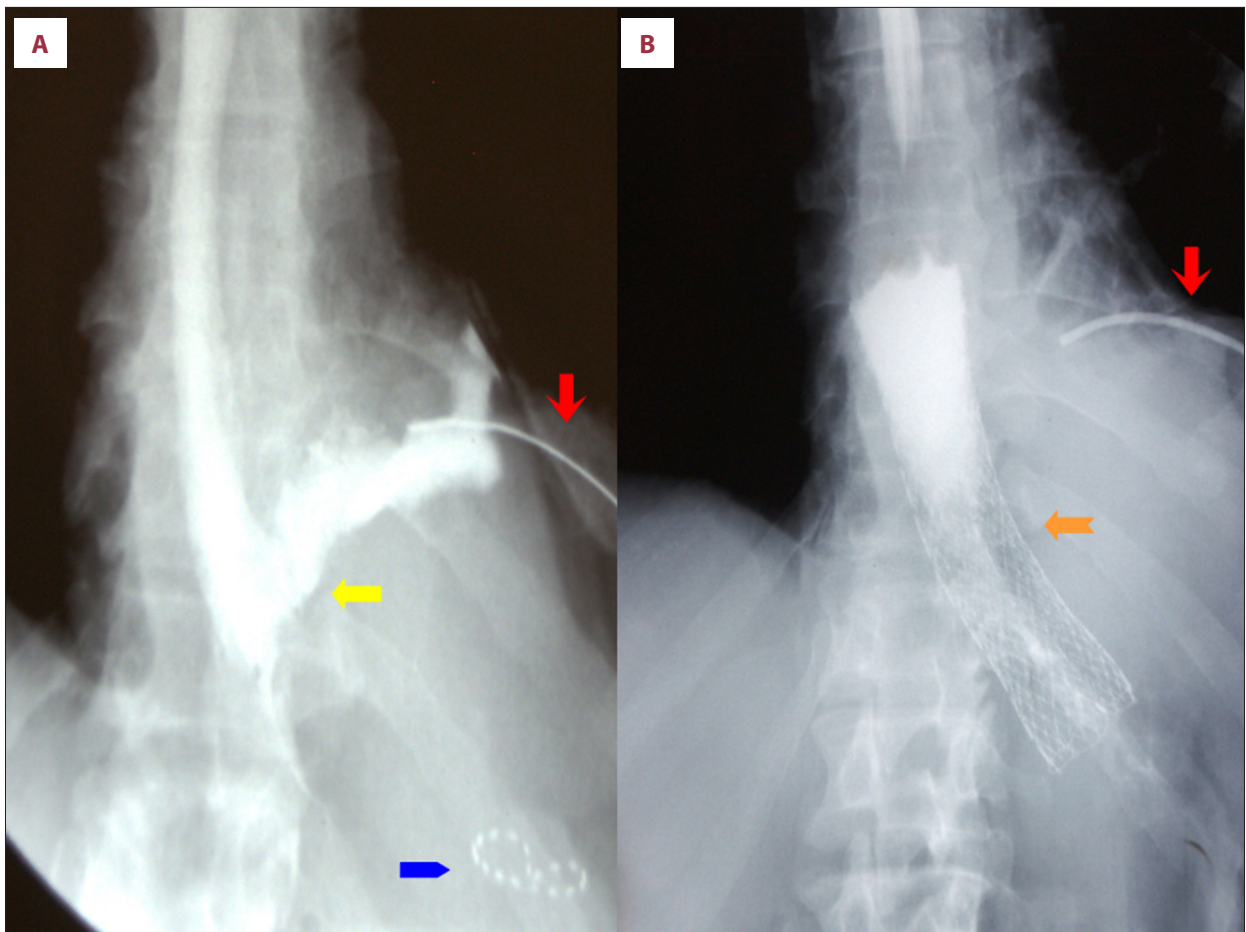


Figure 2. Radiological presentation of esophageal perforation in Boerhaave syndrome and its sealing with a self-expandable metallic stent. **(A)** Demonstrates an extravasation of water-soluble contrast through the esophageal perforation 2 days after endoscopic implantation of the Polyflex stent that migrated to the stomach (yellow regular arrow indicates the site of perforation; blue smooth arrow indicates migrated Polyflex stent; red sharp arrow indicates a chest tube). **(B)** Demonstrates a water-soluble contrast swallow 7 days after endoscopic implantation of the EndoMAXX stent (orange sharp-ended arrow indicates stent across esophago-gastric junction sealing esophageal perforation; red sharp arrow indicates a chest tube).

identified). The patient was still in stable condition with mild dyspnea but not requiring any respiratory support. Endoscopy was performed to reassess the magnitude of esophageal disruption. A 3-cm connection to the pleural cavity with no pre-existing pathology and no specific signs of healing was found (**Figure 1A, 1B**). Endoscopic stent-grafting with a Polyflex 18-23 mm/120 mm (Boston Scientific, MA, USA) self-expandable plastic stent-graft was performed under general anesthesia. Two days later, a water-soluble contrast swallow revealed stent-graft migration to the stomach with persistent fistula to the left pleural cavity (**Figure 2A**). The patient was referred to another session of endoscopic stent-grafting with an EndoMAXX 23-28 mm/120 mm (Merit Medical Systems, UT, USA) fully covered self-expanding metallic stent-graft (**Figure 1C**). The EndoMAXX stent-graft chosen for restenting was wider than the initial plastic stent-graft and was equipped with anti-migration struts. After restenting, the amount of purulent discharge

evacuated from the pleural cavity was gradually decreasing and active chest drainage was switched to gravity drainage 10 days later. A water-soluble contrast swallow performed 7 days after restenting confirmed the proper position of the EndoMAXX stent-graft across the esophago-gastric junction and no extravasation of contrast beyond the esophageal lumen (**Figure 2B**). Subsequently, liquid and then soft diet were resumed. Targeted antibiotic therapy and parenteral nutritional support were administered as a concomitant treatment. The patient was discharged from the hospital 15 days after restenting and 28 days after spontaneous perforation. Two weeks later, the chest drain was empty and fully expanded lungs with no pleural effusion were found on chest X-ray. The chest tube was removed. Three weeks later, a check-up chest X-ray revealed the stent-graft underneath the diaphragm. The patient was asymptomatic. During endoscopy (8 weeks after restenting) we found the site of previous esophageal perforation

sealed with a linear scar and the stent-graft migrated to the stomach (**Figure 1D**). The stent-graft was removed without any complication using a polypectomy snare. Five months after stent-grafting, the patient remained in excellent condition with no esophageal dysfunction.

Discussion

Spontaneous esophageal perforation is a rare esophageal pathology. The rarity of this condition results in very scarce data even in specialist centers and the level of evidence is limited to retrospective case series and expert opinions [12,13]. No consensus has been reached on the most suitable treatment. A turning point in the management of Boerhaave syndrome was the introduction of surgical therapy by Barrett in 1947 [2-6]. Until this time, Boerhaave syndrome was considered fatal. Surgical treatment dramatically reduced mortality. Thus, a surgical approach including primary closure with or without omentoplasty, cervical exclusion, or esophagectomy has traditionally become the treatment of choice [1-6].

In a large meta-analysis including 389 patients with spontaneous rupture, the mortality rate of 14.8% was higher than in all 2971 patients with esophageal perforation – 11.9% [1]. The mortality rate after esophageal perforation was strongly related to the time and type of treatment. If a treatment was applied within 24 hours, the mortality rate was lower than in patients treated later than 24 hours after perforation: 7.4% compared to 20.3%, respectively [1]. Primary repair was associated with a pooled mortality rate of 9.5%, esophagectomy 13.8%, T-tube or any other tube repair 20.0%, and stent-grafting 7.3% [1].

Although surgery remains the main treatment in patients with spontaneous esophageal rupture, endoscopic temporary stent-grafting with fully (FSEMS) or partially (PSEMS) covered self-expanding metal stent-grafts or self-expanding plastic stent-grafts (SEPS) has been proposed recently in the management of esophageal perforation [6-11]. The pooled experience in the treatment of benign esophageal ruptures (mainly iatrogenic) and anastomotic leaks has been reported by van Boeckel based on the systematic review of 25 studies evaluating 267 patients, including 46 (17%) patients with Boerhaave syndrome [8]. Clinical success was achieved in 85% of all patients and was not significantly different among SEPS (84%), FSEMS (85%), and PSEMS (86%) [8]. Stent-graft migration occurred more frequently with SEPS (31%) than FSEMS (26%) or PSEMS (12%) [8]. In the largest case series of 88 patients with upper gastrointestinal leaks or perforations, including 4 patients with Boerhaave syndrome, Swinnen et al reported a clinical success with complete closure of a leak or perforation after endoscopic stent-grafting (exclusively PSEMS) in 59 patients (77.6%), whereas 17 patients (22.4%) had a persistent leak [9]. The

mortality rate was 10.2% [9]. The success rate was directly related to duration of perforation. Immediate closure of a perforation resulted in a success rate of 100% compared with 50% when stent-grafting was applied more than 1 month after the perforation [9]. The success rate was also associated with the degree of contamination. Endoscopic stent-grafting in a non-infectious patient yielded better results (100%) than in chronic leaks (40%) or in infectious patients (75.9%) [9]. Migration of a stent-graft occurred in 12 patients (13.6%) and dysphagia related to tissue hyperplasia occurred in 16 patients (18.2%) [9].

We report a patient with spontaneous esophageal rupture successfully treated by late endoscopic stent-grafting. After initial non-effective conservative treatment, endoscopic stent-grafting was performed 7 days from the onset of symptoms. SEPS (Polyflex) used initially migrated very early to the stomach. SEPS was then replaced by FSEMS (EndoMAXX), which was wider and equipped with anti-migration struts. Implantation of the EndoMAXX stent-graft resulted in clinical success with closure of the esophageal rupture. Two key factors allowing less invasive treatment with endoscopic stent-grafting were adequate drainage of septic contamination from the chest and stable patient condition without general septic complications. The diagnosis of spontaneous esophageal rupture is frequently delayed, probably reflecting the rarity and diverse presentation of this condition. Surgery remains the mainstay of treatment for those who present early with widespread septic contamination. Application of minimally invasive access may further reduce the surgery-related risk and improve outcome [14,15]. In case of widespread septic contamination, it has been reported by some authors that a stent-graft can prevent adequate drainage of purulent discharge, delay healing due to devitalized borders of the esophageal lesion, and potentially erode adjacent structures by constant compression [4]. Thus, they strongly recommend avoiding endoscopic stent-grafting in Boerhaave syndrome [4]. Nevertheless, endoscopic stent-grafting may reduce the need for major surgical operations, including esophagectomy. Thus, it may be an attractive option in patients in poor condition because it allows the preservation of the esophagus in most patients [11]. The majority of experience with stent-grafting for esophageal full-thickness injury has been reported for iatrogenic perforations and anastomotic leaks, when early detection and treatment of well-contained perforations prevent considerable chemical and bacterial contamination and spreading of septic complications [8-10]. Patients with spontaneous esophageal rupture were not discussed separately in these reports, but the delay of endoscopic stent-grafting and considerable infectious contamination resulted in the decrease of success rate down to 40-50% [10]. Another endoscopic alternative option in the management of esophageal perforation is endoscopic vacuum therapy. The results of this therapeutic method are promising, but data on its application in Boerhaave syndrome are scarce [16,17].

Conclusions

One of the most difficult dilemmas in the treatment of Boerhaave syndrome is selection of the most appropriate management for late perforations with severe septic complications. In this situation, an aggressive surgical approach with esophagectomy and immediate or postponed reconstruction has been recommended in large series [3,4]. Although surgery remains the main treatment of Boerhaave syndrome both in early and late diagnosis, an attempt to preserve the esophagus could be considered on an individual basis. Our case indicates that even late after onset of spontaneous esophageal rupture, less invasive treatment by endoscopic stent-grafting with adequate drainage of septic contamination may be an attractive option, preserving esophagus in selected patients

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Conflict of Interest

None.

Declaration of Figures Authenticity

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