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Endoscopic Management of Boerhaave Syndrome: Are Outcomes Better Than Surgery? A Case Report and Review of Literature

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Abstract

Boerhaave syndrome (BS) is a rare clinical diagnosis associated with a high morbidity and mortality rate. Diagnosis of this condition is usually delayed which can lead to a very poor outcome. The timing of presentation and time to management plays a very important role in the prognosis and selection of the management method. With the advances seen in therapeutic endoscopy, many authors have been exploring the possibility of shifting the focus of management from surgery to interventional endoscopy. We present a case report of a patient presenting with BS that was successfully managed endoscopically. We also reviewed the literature on how surgical management compares to endoscopic management and attempted to establish general recommendations from available literature on management of BS.

Keywords: Boerhaave syndrome, Esophageal perforation, Cyclic vomiting syndrome, Endoscopic stent, Endoscopic clip placement

1. Introduction

Boerhaave syndrome (BS) is an effort rupture of the esophagus, which although rare, can be a life-threatening condition with a high mortality.^{1,2} It is unrelated to foreign bodies, instrumentation, trauma or surgery and usually occurs with straining or vomiting where there are increased intra-esophageal pressures.³ Ruptures most commonly involve the left posterolateral wall of the lower 1/3rd of the esophagus but can occur in any segment.⁴ Intra-thoracic rupture of the esophagus can lead to contamination of the mediastinum causing mediastinitis, empyema, pericarditis and rarely sepsis.⁵ Mortality rates with esophageal perforation can amount to up to 35 %.⁶

Clinical presentation can take more than an hour to develop and may only be present in 14 % of

patients.⁴ Hence, diagnosis can be challenging and can be delayed leading to rapid deterioration of the clinical course. An esophagram may reveal extravasation that can aid diagnosis and localize the site of rupture. Computed tomography (CT) can also be done if an esophagram is not feasible.

Management can be either surgical, endoscopic or conservative. In uncomplicated cases where the leak is contained, conservative management may suffice.⁷ For several years surgery, either open or minimally invasive, was the only option.⁸ In the past decade, endoscopic therapy using stents or clips has been increasingly used to manage esophageal perforations.

We present a case of BS from cyclical vomiting syndrome (CVS) that was managed with endoscopic placement of a self-expanding metallic stent (SEMS) with excellent outcomes.

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2. Case presentation

A 38-year-old male with cannabis and poly-substance abuse presented to the emergency room with vomiting and retrosternal chest pain for 2 days. His vital signs were normal and physical exam only revealed a tender epigastric region without any guarding or rigidity. Laboratory workup only revealed mild leukocytosis.

CT chest revealed pneumomediastinum extending into the soft tissues of the neck (Fig. 1A & B) and an esophagram showed active extravasation of gastrograffin from the distal subdiaphragmatic esophagus immediately proximal to the gastroesophageal junction confirming BS (Fig. 1C).

Gastroenterology and surgery were consulted. Esophagogastroduodenoscopy (EGD) revealed a medium-sized nonbleeding perforation at the gastroesophageal junction with adjacent mucosal ulceration. The tissue edges were approximated, and two 11 mm over-the-scope (OTC) clips were placed (Fig. 2A). A fully covered SEMS measuring was placed from the mid-esophagus extending 3 cm distal to the gastroesophageal junction overlapping the site of the perforation (Fig. 2B). The distal end of the stent was left free in the stomach. Two OTC clips were used to secure the proximal end of the stent to the mid-esophageal wall.

An esophagram 24 h later did not reveal any active contrast extravasation. Chest x-ray showed that the pneumomediastinum had resolved. Diet was introduced after 24 h. He remained asymptomatic and hemodynamically stable and was discharged after two days. The patient returned to the emergency room 2 weeks later with vomiting and abdominal pain. CT revealed that the stent had displaced into the stomach. There were no features of pneumomediastinum and esophagogram did not reveal any active extravasation. An EGD was done to extract the stent. The patient had an uneventful hospitalization and was discharged subsequently.

3. Discussion

BS remains particularly challenging since diagnosis can go unnoticed and hence be delayed. A uniform consensus across all published literature is that a delay in intervention is associated with poor prognosis. Given the heterogeneity of study populations and existence of only retrospective studies, it is difficult to determine the best intervention for esophageal perforations. Success of any approach is dependent on carefully selecting patients.^{9,10}

The case presented showed a successful outcome where we performed a closure with OTC clips, followed by placement of SEMS less than 24 h after

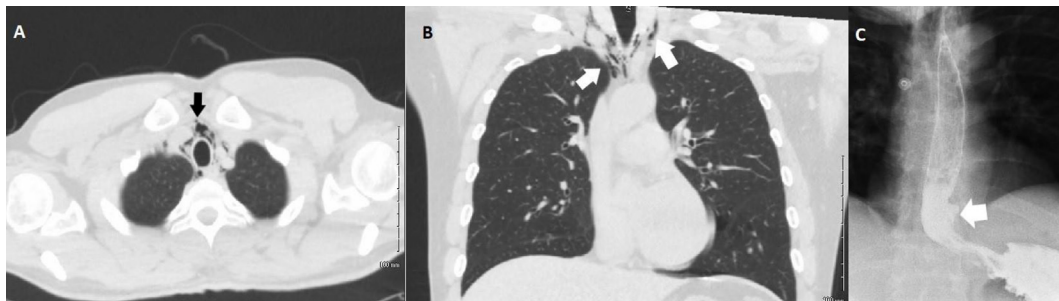


Fig. 1. Radiological images confirming esophageal perforation. A & B depict the pneumomediastinum in the soft tissues of the neck on CT (Arrows). C depicts the esophagram with contrast extravasation from the distal esophagus (White Arrow).

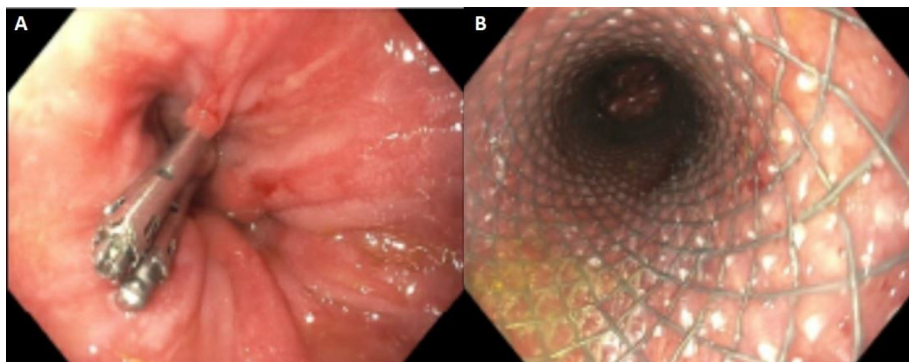


Fig. 2. Endoscopic images of stent placement. 2 A shows the two clips to close the defect. 2 B shows the placement of the esophageal stent.

presentation. The postprocedural esophagogram did not reveal any active extravasation of contrast confirming successful closure preventing any contamination of the mediastinum. The repeat esophagogram where the patient presented with stent migration confirmed that the perforation had healed.

Objectives of the treatment for BS should include closure of the site of perforation, prevention of extravasation of the contaminants into the mediastinal and pleural spaces and if present, removal of any foci of infection from these spaces. Infrequent presentation and delayed diagnosis of BS contribute to the lack of comparative studies between surgical and endoscopic management. 10-year studies of patients with spontaneous esophageal perforations comparing surgery and endoscopic stent placement showed lower mortality rates and shorter hospital stays but more readmission rates with the latter.¹¹ Surgical options for esophageal perforations include open or laparoscopic repair or esophagectomy with diversion procedures. Mortality rates with surgery widely range from 9.5 % to 36 % and doubles if treatment is delayed greater than 24 h.^{9,11,12} Mortality rates were higher since some of the study population also included intervention for patients with malignant esophageal perforations. In one study, 85 % of patients who had undergone a stent placement required subsequent procedures such as drainage of empyemas.¹³

Esophageal stents have been historically used as palliative therapy for esophageal malignancies.¹⁴ Continued advancements, increased availability and favorable outcomes have encouraged their use for treating esophageal perforations. Endoscopic management includes placement of OTCs, through-the-scope clips, partial or fully covered SEMs. Newer methods of closure which have been less studied in esophageal perforations include endoscopic suturing, vacuum-assisted closure or using sealants such as fibrin glue or cyanoacrylate to fill defects. Stents are usually recommended for rupture up to 70 % of the esophageal circumference, while surgery is usually reserved for larger leaks and ruptures.^{15,16} No differences in clinical success rates have been seen between plastic and metallic stents.¹⁷ Clip placements are more successful if patients remain stable and there are no signs of infection or contamination. Despite a success rate of 76 % in some patients, over 75 % of them may require subsequent surgery in case of clip failure.¹⁸ Delayed intervention with clip placement has a higher chance of failure since the site of perforation may be more inflamed and necrotic which may prevent the clip from holding in place. Proximal lesions

extending up to the middle of the esophagus are more technically challenging for clip placement.^{19,20} Overall, better outcomes are seen with clip placement if the size of the perforation is around 10–20 mm.¹⁸ We took a combined approach of placing a clip to close the perforation with subsequent placement of a metallic stent.

In comparison to surgical management, stent placements have more successful outcomes with mortality rates ranging from 7.3 to 9%.^{11,16,21} Additional advantages include quicker resumption of an oral diet, shorter healing times and critical care durations.^{16,21} Studies have shown that by placement of a stent, at least 80 % of the perforations can be sealed successfully.^{22,23} Timing of intervention plays an important role in the prognosis, with higher rates of mortality seen in delay greater than 24 h.^{13,24} Endoscopic management is especially useful in salvage therapy after surgical management fails and in patients too sick for surgery.⁹ Overall, endoscopy with either placement of stents only or a dual approach therapy with stents and clip placement, has shown to have lower mortality rates while at the same time showed excellent outcomes for patients with any type of esophageal perforation. Endoscopic suturing can also be used in the primary repair of perforations to anchor esophageal stents. Endoscopic vacuum-assisted closure has also been shown to have great success in esophageal leakage but has not been studied in esophageal perforations. It is important to remember that endoscopy is more successful if there is no contamination of the mediastinal and pleural space or if the source of infection is controlled by percutaneous drainage, laparotomy or video-assisted thoracoscopic surgery (VATS).

10-year studies have shown that the highest rates of stent failure were seen with BS. Stent migrations, erosion of the mucosa, bleeding and stent perforation are some of the risks encountered with stent placement. Migration rates range from 10 to 37 %.^{22,25,26} Hence anchoring the stent is important. We anchored the stent in our case with an over-the-scope clip, but this did not prevent stent migration. Stent removal is recommended in 5–6 weeks. With PSEMS, tissue overgrowth can make extraction difficult, but can also decrease stent migration rates.²⁵

Conservative approaches also remain an option for the management, with some studies reporting better results than surgical or endoscopic interventions in terms of length of stay without any difference in terms of mortality.^{27–29} This was in spite of the median size of the perforations being 20 mm which was closely matched to sizes in the

Table 1. Management recommendations from literature review.

1	Treatment options are dependent upon timing of presentation, size of the perforation, location of the perforation and extent of contamination, absence of sepsis along with prognosis of the patient.
2	Patients who were managed less than 24 hours after the perforation with any treatment modality have a far better outcome.
3	A difference between surgical or endoscopic management is difficult to determine. Endoscopy remains minimally invasive and will avoid surgical complications
4	If there are signs of mediastinal or pleural contamination, stenting can be an option if coupled with drainage procedures such as chest tube drainage. ¹⁰
5	Esophageal stenting can also be an option for salvage therapy after failure of surgery. ⁹
6	If perforation is less than 1 cm and presentation is less than 24 hours, endoscopic therapy can be done with clips or sutures. If there are signs of extravasation, drainage needs to be done. If the size of the perforation is greater than 1 cm, stents will be required. ⁹
7	Conservative management can be attempted if perforations are intramural and there are no signs of pleural or mediastinal contamination, or the pleural effusions are drained or contained.

surgical and endoscopic group.²⁹ Better outcomes can be seen especially if there are no signs of contamination or if the effusions are well drained.³⁰ Most of the other studies remain biased since conservative management is vastly attempted in small perforations or if the patient is unfit for surgery. Conservative management can delay surgical management if required later which by that time can worsen the prognosis since patients would usually fall in the greater than 24 h category. The esophageal perforation severity scoring developed by the Pittsburgh group used in a retrospective analysis showed patient's with the lower score had better outcomes if they were managed conservatively.³¹ Table 1 depicts the overall management recommendations of Boerhaave Syndrome that has been summarized via a literature review.

4. Conclusion

BS remains a grave condition with a high mortality rate. Delayed management is associated with poor outcomes. Given its rare presentation in the clinical setting and the heterogeneity of the study populations, it is difficult to adequately compare all the modes of treatment. Results of endoscopic intervention with stents and clip placement are comparable to surgical methods, with the added advantage of avoiding risks associated with surgery. More trials are required which would help us determine whether endoscopic interventions can replace the role of surgical intervention.

Disclaimers

All the authors confirm that this manuscript exhibits original research that has not been previously published or presented at any conferences. It is not under consideration elsewhere.

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Conflict of interest

None of the authors have any conflicts of interest.

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