Case Report: Esophageal Necrosis and Perforation Presenting as Intractable Hiccups

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INTRODUCTION

Esophageal necrosis is a rare disorder with less than 200 reported cases in the literature and a mortality rate of 1 in 3. Patients typically present with symptoms of hematemesis and melena.

CASE PRESENTATION

A 52-year-old male with a history of alcohol abuse, LA grade D esophagitis, hiatal hernia, and chronic hiccups (managed with baclofen, pantoprazole, and thorazine) presented with 5 days of intractable hiccups attributed to running out of thorazine. He presented febrile to 38.1C, tachycardic to 120s, with a leukocytosis to 13, lactate of 5.8, and an acute kidney injury. A CT of the abdomen showed a hiatal hernia with thickening of the lower esophagus and edematous stomach with peri-gastric pneumoperitoneum. An emergent exploratory laparotomy showed no evidence of gastric perforation, a negative air leak test (via nasogastric tube at 40cm and 55cm), and no initially obvious distal esophageal defect. Intraoperative EGD found circumferential esophageal necrosis spanning 25 to 40cm from the incisors with an anterior 1-cm mural defect abutting the GE junction. A covered metal esophageal stent was placed by gastroenterology and reinforced with an omental patch by general surgery. A nasogastric tube was left to drain the stomach and a feeding jejunostomy tube was placed in anticipation of prolonged NPO status. Wide drainage was achieved with a JP drain and bilateral chest tubes. Antibiotics and antifungals were continued for two-weeks. At 4 weeks post-operatively, an EGD showed a healed perforation with improving esophagitis but with stenosis at 25 to 28cm, requiring replacement of a covered stent. At 11 weeks, repeat EGD showed significant fibrosis and stenosis distal to the previous stent, requiring addition of a second stent. 6 months after the index procedure, the patient still demonstrates the presence of an esophageal stricture requiring stenting.

RELEVANT IMAGES

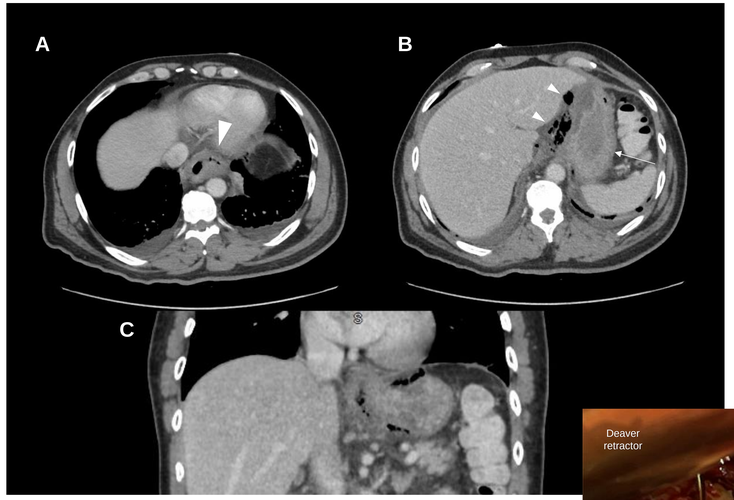


Figure 1. CT abdomen/pelvis obtained on presentation to the emergency department.

A. Axial image of thorax demonstrating pneumo-mediastinum around the esophagus (white arrowhead).

B. Axial image of upper abdomen showing air within the lesser sac (white arrowheads). Also seen on this slice is the thickened stomach wall (white arrow).

C. Coronal image also demonstrating pneumo-peritoneum around the stomach (white arrowheads).

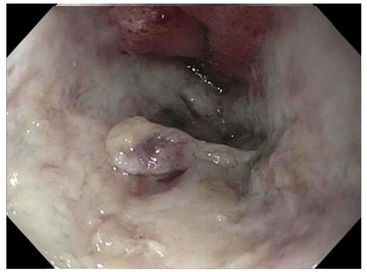


Figure 2. Intraoperative upper endoscopy.

Circumferential esophageal necrosis seen at the patient's index procedure starting from 25-cm to 40-cm from the incisors.

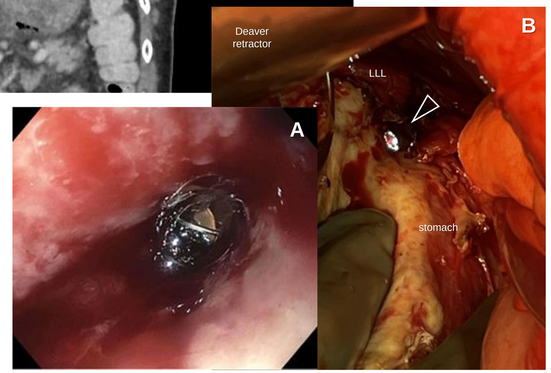


Figure 3. Identification of esophageal perforation during laparotomy with confirmation on endoscopy.

A. Endoscopic view of esophageal perforation showing a ~1-cm defect with visible empty ring clamp. **B**. View of perforation at GE junction (white arrowhead outline). LLL: left lobe of liver.

POST-OPERATIVE ENDOSCOPIES

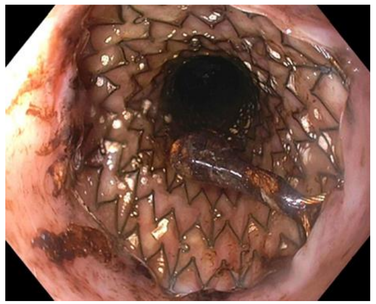


Figure 4. 4-week post-surgical endoscopy showing stent in place.

Moderate diffuse mucosal inflammation (LA Grade C esophagitis) seen in the middle esophagus 1-month after index procedure. This stent was replaced with a fully covered 23mm x 100mm stent under fluoroscopic guidance.



Figure 5. 6-month post-surgical endoscopy demonstrating mid-esophageal stricture.

Even at 6-months, a persistent stricture is seen from 30-cm to 42-cm requiring repeated stenting with covered 12mm x 120mm stent.

DISCUSSION

The etiology of esophageal necrosis is controversial and multifactorial, likely involving an acute triggering event in the setting of chronic comorbidities such as reflux, general debilitation, or ischemia. Management is primarily supportive with treatment of underlying conditions. In the case of perforation, prior case reports show survival after VATS repair or esophagectomies. This case illustrates a unique presentation of intractable hiccups, with esophageal perforation successfully managed by endoscopic stenting and wide drainage.

CONCLUSIONS

In the rare case of esophageal necrosis leading to perforation at the GE junction, patients may be managed with a combination of buttressed repair with omental patching along with endoscopic stenting. Wide drainage allowed for surveillance of the repair in the immediate post-operative period while regular endoscopies monitor for later post-operative complications including stricture.

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