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CASE REPORT | LIVER

Massive Gastrointestinal Hemorrhage Due to an Arterioenteric Fistula From a Hepatic Artery Pseudoaneurysm

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ABSTRACT

Hepatic artery pseudoaneurysms are a rare cause of upper gastrointestinal bleeding with a high mortality rate. We report a case of a 37-year-old woman who presented with massive gastrointestinal hemorrhage and was found to have an arterioenteric fistula from a hepatic artery pseudoaneurysm. Esophagogastroduodenoscopy revealed a 1.5-cm ulcer overlying a large mucosal bulge that compressed the lumen of the duodenal bulb. A vascular stent graft was placed successfully over the pseudoaneurysm neck. We report the first case of the unique intraluminal appearance of an enteric fistula related to a hepatic artery pseudoaneurysm.

INTRODUCTION

A hepatic artery pseudoaneurysm (HAPA) is a rare cause of upper gastrointestinal bleeding (UGIB). A pseudoaneurysm is a contained hematoma that forms due to a defect in the arterial wall.[1] A HAPA is commonly caused by blunt abdominal trauma or occur iatrogenically after laparoscopic surgeries or interventional radiologic procedures.[2,3] Chronic pancreatitis is also thought to cause HAPAs.[4] The risk of rupture of all hepatic artery aneurysms (true aneurysms and pseudoaneurysms) has been reported to be 20–80%, with mortality rates up to 43%.[2,5]

Although many patients with a HAPA are asymptomatic, the most common symptoms include right upper quadrant pain and gastrointestinal bleeding. Patients may present with acute abdomen and shock due to intraperitoneal hemorrhage, or with UGIB due to an arterioenteric fistula. Rarely, biliary obstruction can result from an aneurysm compressing the biliary tree.[6] It is essential for the practicing gastroenterologist to consider a hepatic arterioenteric fistula in the differential diagnosis of UGIB because definitive management of the underlying aneurysm is key to the patient's survival.

CASE REPORT

A 37-year-old woman with a history of alcohol abuse presented to her local hospital with 1 day of hematochezia and hematemesis. She was found to be in hemorrhagic shock with blood pressure 74/31 mm Hg, pulse 141 beats/min, hemoglobin 6.3 gm/dL, platelets 142,000/mL, and international normalized ratio 1.3. She required 10 units of red blood cell transfusions in the first 24 hours. Esophagogastroduodenoscopy (EGD) revealed a 1.5-cm ulcer with a visible vessel in the duodenal bulb, which was treated with bipolar cautery. The ulcer was overlying a large mucosal bulge that compressed the lumen of the duodenum.

By hospital day 3, the patient was hemodynamically stable and required no further transfusions. A computed tomography angiography of the abdomen revealed a right HAPA that was compressing the duodenum (Figure 1). She was transferred to a tertiary care medical center on hospital day 6 for further management.

Upon transfer, the patient was hemodynamically stable without signs of active UGIB, with hemoglobin 9 gm/dL, platelets 138,000/mL, and international normalized ratio 1.2. After multidisciplinary consult, the decision was made

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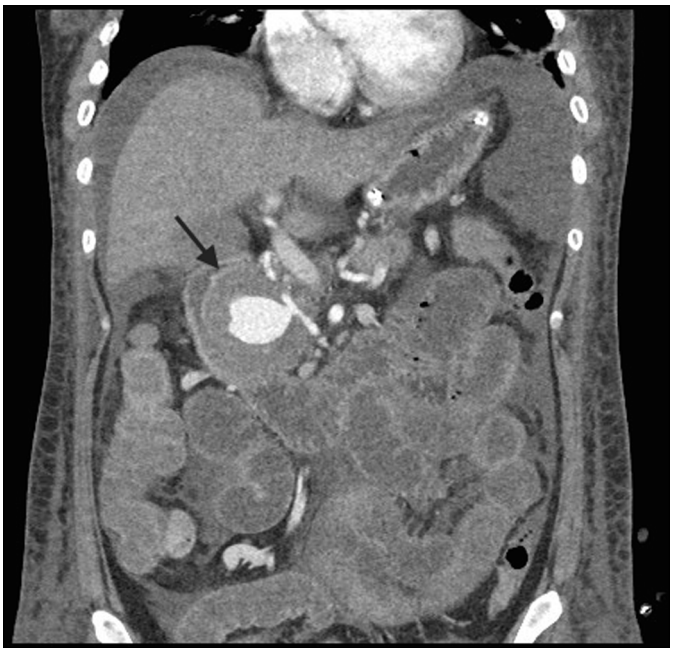

Figure 1. Abdominal computed tomography angiography showing a 5.2-cm right hepatic artery pseudoaneurysm compressing the duodenum.

to proceed with endovascular stenting of the right hepatic artery. Prior to endovascular repair, the patient acutely decompensated with hematemesis, hypotension, and subsequent loss of pulses due to hemorrhagic shock. After 15 minutes of cardiopulmonary resuscitation, she regained her pulse. She then proceeded with endovascular stenting given that it was the best chance for survival. A 5 mm x 5 cm covered stent graft was placed successfully over the pseudoaneurysm neck (Figure 2).

By the next day (hospital day 7), the patient was hemodynamically stable and required no further transfusions. On hospital day 10, she had an episode of melena (first bowel movement since stenting) without change in hemodynamics or hemoglobin. Repeat angiography demonstrated no leak from the stent. EGD revealed no active bleeding, but a bulge in the duodenal bulb with 2 fistulous openings outlined by clean-based ulcers that communicated with the pseudoaneurysm (Figure 3). The remainder of the hospital course was uneventful, and she was discharged on hospital day 14.

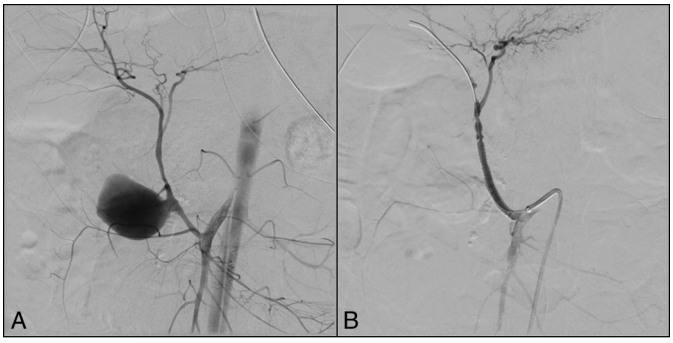

Figure 2. (A) Pre-stenting angiogram showing filling of the pseudoaneurysm with contrast. (B) Post-stenting angiogram showing no further extravasation into the pseudoaneurysm.

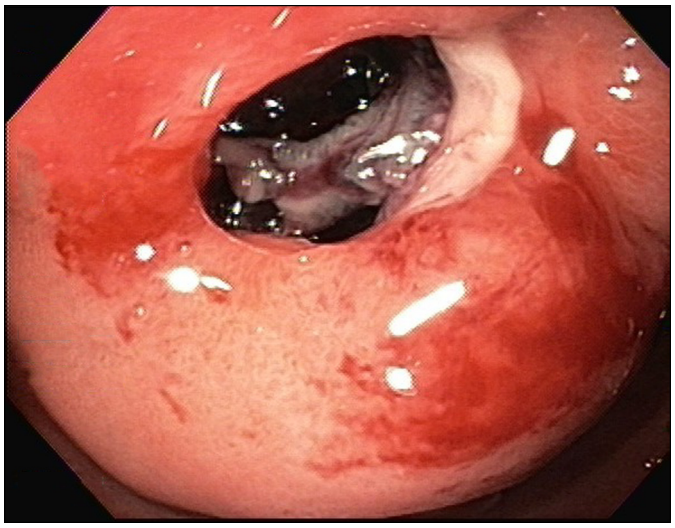

Figure 3. Esophagogastroduodenoscopy showing no active bleeding but a bulge in the duodenal bulb with 2 fistulous openings outlined by clean-based ulcers that communicated with the pseudoaneurysm.

DISCUSSION

Although typically asymptomatic, HAPAs can fistulize into the duodenum, resulting in UGIB. A possible mechanism by which fistulization occurs is the constant pressure and pulsation exerted externally by the aneurysm on the bowel wall, leading to ischemia and ulceration into the lumen.[7] Similar to cases of aortoenteric fistula, patients may present with a herald bleed, which is a smaller bleed that precedes a catastrophic bleed. The typical interval between the herald bleed and subsequent catastrophic hemorrhage is unknown, but a lag time of 6 days has been reported for aortoenteric fistulas.[8] In our patient's case, there was a period of 5 days between the bleeding episodes. It is clear that treating the aneurysm in this period between the herald and catastrophic bleed is critical for survival. A review of 4 case reports reveals that patients who were treated during the interval period survived, whereas likelihood of survival is very low once the catastrophic bleeding occurs.[2,3,9,10]

Diagnosis of HAPA is made by imaging, including computed tomography angiography and direct angiography.[9] Endoscopy is not considered a mode of diagnosis, although it is likely that the gastroenterologist will be called when a patient presents with hematemesis from an enteric fistula. There are typical characteristics that may be encountered on endoscopy that may lead an endoscopist to consider an underlying

HAPA. Most notably, a duodenal bulb bulge may be seen immediately upon passing the pylorus.[3,9] The bulge is created by the aneurysm compressing the duodenum externally. A bleeding ulcer overlying the bulge may also be encountered.

Regardless of the size, HAPAs are treated when found due to the high risk of rupture.[1,5] Traditionally, surgery has been the definitive treatment. Common techniques include ligation, venous or synthetic grafting, and hepatic resection.[5,11] More recently, endovascular repair with coil/glue embolization and stent grafts have been employed with success and low postprocedural morbidity.[11,12] Higher rates of stent infection have been reported in aortic grafts complicated by aortoenteric fistulas, so prophylactic antibiotics should be considered.[13]

Two potential etiologies can be considered as the cause of our patient's HAPA. She was in a significant car accident when she was a child, and delayed presentation of HAPA after blunt abdominal trauma has been reported.[3,9] Another potential cause is subclinical pancreatitis, which can result in pseudoaneurysm formation given her history of alcohol.

In conclusion, we present a patient with catastrophic upper gastrointestinal hemorrhage due to a HAPA with arterioenteric fistula, which was treated with endovascular stent graft. Remarkably, our patient survived without any end organ damage after a pulseless arrest due to the massive hemorrhage. We also report for the first time the unique intraluminal appearance of the enteric fistula related to a HAPA. It is critical to definitively identify and manage such an aneurysm expeditiously after the herald bleed, and prior to the ensuing catastrophic bleed, to reduce morbidity and mortality.

DISCLOSURES

Author contributions: N. Takatori wrote the manuscript and is the article guarantor. D. Patel wrote and edited the manuscript.

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Informed consent was obtained for this case report.

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