A rare case report of acute upper gastrointestinal hemorrhage due to splenic artery pseudoaneurysm

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Abstract
Splenic artery aneurysm and splenic artery pseudoaneurysm are rare vascular pathologies. The splenic artery represents the third most common site for intra-abdominal aneurysms. In contrast with true splenic artery aneurysm, splenic artery pseudoaneurysm is typically symptomatic, presenting with a range of symptoms, from abdominal pain to hemodynamic instability due to rupture. However, gastrointestinal hemorrhage is an uncommon complication of splenic artery pseudoaneurysm. We report a case of acute upper gastrointestinal hemorrhage due to splenic artery pseudoaneurysm rupture. The patient was successfully treated by endovascular intervention.

Keywords
Upper gastrointestinal hemorrhage, splenic pseudoaneurysm, endovascular intervention

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Introduction
First described by Beaussier in 1770 at necropsy,1 splenic artery aneurysms (SAAs) are uncommon lesions that are more frequently detected due to advancements in modern medical techniques, particularly cross-sectional imaging modalities. The true prevalence of SAA varies widely, from 0.098% to 10.4% based on autopsy studies.2–4 Splenic artery pseudoaneurysm (SAP) is an even rarer event. In a review of 37 visceral pseudoaneurysms treated at the Mayo Clinic over an 18-year period, only 10 cases of SAP were identified,5 and fewer than 200 cases have been reported in the English literature to date.

In contrast with true SAA, SAP is often symptomatic, and patients often exhibit abdominal pain and signs of hypovolemic shock due to intraperitoneal hemorrhage. Upper gastrointestinal (GI) hemorrhage is an uncommon complication of SAP, with only a few cases reported in the literature. We present a case of SAP rupture that caused a massive upper GI hemorrhage. This patient was successfully treated by endovascular intervention. Moreover, we summarize this vascular pathology in a relevant literature review to highlight the importance of clinical suspicion for accurately diagnosing SAP in patients with upper GI bleeding.

Case description
A 50-year-old man was admitted to the emergency department due to hematemesis on the same day. The patient reported vomiting twice, producing approximately 200 mL of dark red blood containing clots. In addition, the patient described melena once on the presentation day. The patient experienced dizziness and was quickly transferred to the hospital. The patient’s history revealed diabetes with no previous surgeries or trauma and no records of cirrhosis or previous episodes of gastrointestinal bleeding.

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GI hemorrhage. In addition, the patient denied any history of tobacco or alcohol abuse. Upon arrival at the emergency department, the patient was pale, with a weak peripheral pulse, tachycardia (130 bpm), and fever (40°C). The patient's blood pressure was initially slightly high (140/90 mm Hg) but abruptly decreased to 90/60 mm Hg. A physical examination revealed a non-tender abdomen with moderate epigastric pain. The insertion of a nasogastric tube resulted in the drainage of 100 mL red blood. Initial lab tests indicated acute anemia, with a hemoglobin level of 9.8 g/dL and hematocrit at 31.4%. The platelet count (101 K/µL) was slightly low, and the white blood cell count (7.84 K/µL) and the international normalized ratio (INR; 1.25) were normal. The GI department was consulted, and contrast-enhanced computed tomography (CT) was requested to determine whether contrast extravasation could be detected, but not the GI endoscopy since the patient’s condition is unstable. The CT results exhibited a pseudoaneurysm at the splenic hilum, pressed into the posterior of the stomach and bulging to the fundus gastric mucosa, which measured 17 mm × 19 mm. No active bleeding was noted due to the absence of contrast extravasation.

Numerous intrasplenic hypoattenuating lesions were poorly enhanced on multiple phases, suggesting inflammatory or abscess lesions (Figure 1). In addition, an area of consolidation was observed at the left lower lobe, which might have been pneumonia. The patient was, therefore, referred to an interventional radiologist to perform an endovascular embolization of the SAP.

**Procedures**

Under local anesthesia, a 6F femoral sheath was inserted into the right femoral artery. The celiac trunk was catheterized using a 5F Yashiro catheter (Glidecath, Terumo, Japan). An arteriogram demonstrated a pseudoaneurysm at the upper pole of the splenic hilum without active contrast extravasation. The pseudoaneurysm was approached using a 2.2F coaxial microcatheter system (Progreat, Terumo, Japan). The SAP was embolized with a mixture consisting of 0.5 mL n-butyl cyanoacrylate (NBCA) and 1.5 mL lipiodol (1:3 ratio). The final angiogram showed the complete occlusion of the pseudoaneurysm (Figure 2). For several days following...
the procedure, the patient appeared to recover well, with no further incidents of hematemesis or melena. However, the patient developed sudden respiratory failure 3 days after the embolism and was diagnosed with acute respiratory distress syndrome. The patient’s white blood cell count rose from 7.84 to 13.7 K/µL, and the C-reactive protein level was 188.09 mg/L. The consolidation observed on the left lower lobe during the prior CT developed into bilateral lung consolidation, as revealed by chest X-ray (Figure 3). The patient was transferred to the intensive care unit, treated with a combination of imipenem and metronidazole. Blood culture was negative. Echocardiography showed tachycardia, left atrium dilatation without thrombus, and no signs of bacterial endocarditis. Unfortunately, the patient’s condition got worse, and he died after 7 days of critical care due to severe lung damage.

Discussion

SAP is a rare clinical entity, with fewer than 200 cases reported in English literature.\(^5\) In contrast with SAA, which involves three full layers of a vessel, including the intima, media, and adventitia, SAP only involves two layers, the intima and media. The formation of SAP is primarily attributed to pancreatitis (52%), trauma (29%), iatrogenic postoperative causes (3%), and, rarely, gastric ulcer disease (2%).\(^5,6\) In the present case, the patient had no history of previous pancreatitis episodes or recent traumatic events. Thus, the etiology of the pseudoaneurysm could not be determined.

However, multiple splenic lesions were identified, indicating inflammation, and an area of consolidation area was detected in the left lower lobe. We assume that some association exists between the pseudoaneurysm and these inflammatory lesions. In contrast to SAA, SAP is symptomatic in most cases. A large review of medical literature incidentally detected only 2.5% of the similar cases.\(^5\) The clinical presentation of SAP has been associated with abdominal pain (29.5%), melena (26.2%), hemorrhage into the pancreatic duct (20.3%), and hematemesis (14.8%).\(^5\) The risk of SAP rupture has been estimated at approximately 37%, with a mortality rate of 90% if left untreated.\(^7,8\)

The risk of rupture exists regardless of the pseudoaneurysm size.\(^5\) GI hemorrhage is a remarkably uncommon complication of SAP rupture, posing diagnostic challenges for clinical physicians. A literature search performed on the PubMed database revealed 11 reported cases of GI hemorrhage due to SAP rupture to date.\(^6,7,9–17\) In addition, because an SAP rupture is a life-threatening condition, a diagnosis should be promptly determined based on endoscopy or imaging modalities, especially CT. The fatal complications are independent of pseudoaneurysm size; therefore, treatment planning should occur immediately upon diagnosis. We did not send the patient to a GI endoscopist due to hemodynamic instability in the present case; therefore, CT was a more suitable solution for identifying signs of active extravasation.

Based on a review of 145 cases, the current therapeutic options for SAP include embolization, splenectomy, and distal pancreatectomy (26%); splenectomy alone (11%); ligation alone (10%); endovascular stenting (4%); ligation and splenectomy (3%); distal pancreatectomy alone (2%); total pancreatectomy alone (1%); and splenectomy with total pancreatectomy (1%).\(^5\) The mortality and morbidity risks associated with surgical intervention are 1.3% and 9%, respectively.\(^18\) Therefore, depending on the hemodynamic state of the patients, endovascular interventions, which are minimally invasive and have low procedure-related morbidity and mortality risks, may be ideal first-line treatment options, with a success rate ranging between 75% and 98%.\(^18,19\) The choice of embolic agents varies, depending on experience, personal preference, and availability, and can include coils, embolic particles (embospheres, gel foam, etc.), and liquid embolic agents, such as NBCA.

Conclusion

SAP is an uncommon vascular pathology, and upper GI hemorrhage is a rare complication of SAP rupture. Thus,
acknowledging SAP rupture as a potential etiology for GI hemorrhage is important for identifying this issue. A prompt diagnosis and urgent treatment strategy should be employed to achieve good results. In such cases, CT angiography is the preferred imaging modality to identify the source of bleeding and evaluate splanchnic vasculature abnormalities. Depending on the patient’s hemodynamic stability, endovascular intervention should be considered as a first-line treatment due to its minimal invasiveness. This case highlights the importance of early diagnosis, treatment, and the consideration of SAP as a potential differential diagnosis for upper GI bleeds.

**Author contributions**

N.D.L. and N.M.D. contributed equally to this article as co-first authors. All authors read and approved the final manuscript.

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**Ethical approval**

Our institution does not require ethical approval for reporting individual cases or case series.

**Informed consent**

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**Data availability statement**

All data generated or analyzed during this study are included in this article and/or its supplementary material files. Further enquiries can be directed to the corresponding author (Dr. Nguyen Minh Duc: bsnguyenminhduc@pnt.edu.vn).

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**Figure 3.** (a) The left lower lobe revealed consolidation on X-ray CT at the time of admission (black arrow). (b) The following chest X-ray indicated the development of bilateral consolidation (white arrows).
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