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CASE REPORT

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Massive Upper Gastrointestinal Bleeding from a Splenic Artery Pseudoaneurysm Caused by a Penetrating Gastric Ulcer: Case Report and Review of Literature

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

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Summary

Background: Splenic artery aneurysm and pseudoaneurysm are rare pathologies. True aneurysms are usually asymptomatic. Aneurysm rupture occurring in 2–3% of cases results in bleeding into the lesser sack, peritoneal space or adjacent organs typically presenting as abdominal pain and hemodynamic instability. In contrast, pseudoaneurysms are nearly always symptomatic carrying a high risk of rupture of 37–47% and mortality rate of 90% if untreated. Therefore, prompt diagnosis and treatment are essential in the management of patients with splenic artery pseudoaneurysm. Typical causes include pancreatitis and trauma. Rarely, the rupture of a pseudoaneurysm presents as upper gastrointestinal (UGI) bleeding. Among causes, peptic ulcer is the casuistic one.

Case Report: This report describes a very rare case of recurrent UGI bleeding from a splenic artery pseudoaneurysm caused by a penetrating gastric ulcer. After negative results of endoscopy and ultrasound, the diagnosis was established in CT angiography. The successful treatment consisted of surgical ligation of the bleeding vessel and suture of the ulcer with preservation of the spleen and pancreas, which is rarely tried in such situations.

Conclusions: The most important factor in identifying a ruptured splenic artery pseudoaneurysm as a source of GI bleeding is considering the diagnosis. UGI hemorrhage from splenic artery pseudoaneurysm can have a relapsing course providing false negative results of endoscopy and ultrasound if performed between episodes of active bleeding. In such cases, immediate CT angiography is useful in establishing diagnosis and in application of proper therapy before possible recurrence.

MeSH Keywords: Aneurysm, False • Angiography • Gastrointestinal Hemorrhage • Multidetector Computed Tomography • Splenic Artery

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Background

Aneurysm of the splenic artery is a rare pathology. The first case of splenic artery aneurysm (SAA) was described by Beaussier in 1770 [1]. Since then, this pathology has been detected with greater frequency as a result of increasing use of accurate imaging methods. It is the most commonly diagnosed visceral aneurysm (60%). Prevalence of SAA is estimated to range from 0.01% to 10.4% according to different data [2]. It is four times more common in women. SAA is usually caused by or associated with pancreatitis, pregnancy (not a cause of aneurysm formation but associated with a higher risk of rupture), portal hypertension, fibromuscular dysplasia, vasculitis, liver transplantation, splenomegaly and trauma. Atherosclerosis is a very rare cause of SAA [3]. Clinically, 97.5% of SAA are asymptomatic. The risk of rupture is estimated at 2–3% [2]. Typical clinical presentations of a ruptured SAA include abdominal pain and hemodynamic instability.

Pseudoaneurysm of the splenic artery is even more rare; there have been about 160 cases reported so far. It is most often caused by pancreatitis (52%) or trauma (29%) [4]. Unlike true aneurysms, splenic artery pseudoaneurysms nearly always present with symptoms. In a largest published case series, pseudoaneurysm was found incidentally in only 2.5% [4]. The most common clinical presentations are abdominal pain (29.5%), hematochezia or melena (26.2%), hemorrhage into the pancreatic duct (20.3%) and hematemesis (14.8%) [4]. The risk of rupture of splenic artery pseudoaneurysm can be as high as 37–47%, with the mortality rate approaching 90% when untreated [4]. Furthermore, both small and large pseudoaneurysms are at a similarly high risk of rupture [5]. Therefore, prompt diagnosis and treatment are critical in the management of such patients.

This case report presents an unusual cause of massive upper gastrointestinal (UGI) bleeding from a ruptured splenic artery pseudoaneurysm as a result of erosion by a penetrating gastric ulcer. To our best knowledge, only four cases of the same etiology have been reported in the literature [4,6]. However, the presented case is unique due to the unusual clinical course resembling "double rupture" behavior, which made it diagnostically challenging. The patient was successfully treated with splenic artery ligation and suture of the ulcer with preservation of the spleen and pancreas that is rare in such cases.

Case Report

A previously healthy 57-year-old male presented in the emergency department after an episode of syncope preceded by a sudden, severe abdominal pain while walking in the woods. On admission he was pale, sweaty, with signs of hypovolemic shock (BP of 40/0 mmHg, tachycardia of 105 bpm) and acute anemia (hemoglobin of 6.7 g/dL). Blood clots were detected on rectal examination. He had no significant medical history, particularly of pancreatitis, alcohol abuse, peptic ulcer disease, abdominal trauma or surgery. He was admitted to the surgery department with a suspicion of UGI bleeding.

During the next 48 hours the patient was stabilized with transfusion of 8 units of PRBC and 4 units of FFP. Upper endoscopy performed on the second day after admission did not reveal any pathology, particularly any source of bleeding or blood clots. Lower endoscopy was planned in two or three days. Abdominal ultrasound did not show any abnormalities.

During the next two days the patient improved significantly. He was normotensive with hemoglobin level of 10.6 g/dL. On the fourth day morning he was eager to be discharged. However, after an hour he suddenly collapsed. His BP was 75/50 mmHg and hemoglobin dropped to 9.3 g/dL. In order to localize the source of bleeding, emergent CT angiography was carried out (Figure 1). The key finding was the presence of massive intragastric bleeding from a ruptured distal splenic artery pseudoaneurysm. Contrast extravasation into the stomach was observed, indicating an active hemorrhage. Pseudoaneurysm with a diameter of 0.6 cm was localized on the posterior wall of the stomach in proximity to the cardia and surrounded by a thrombus. Erosion of an adjacent wall of the stomach was visible with necrotic masses filling a 4.0-cm ulcer crater. Additionally, critical stenosis of the celiac artery was found with collateral supply through the pancreaticoduodenal, short gastric and gastroepiploic arteries.

Shortly after CT the patient developed hypovolemic shock. After transfusion of 3 units of PRBC, it was decided to perform an emergency laparotomy. Intraoperatively the stomach was filled with blood clots. A bleeding ulcer of the posterior wall of the stomach, 4.0 cm in diameter, was revealed. A pseudoaneurysm of a branch of the splenic artery was identified as the source of bleeding. Bipolar ligation of the bleeding vessel was performed and the gastric ulcer was sutured. The spleen and pancreas were preserved.

After surgery the patient developed ARDS and was transferred to the intensive care unit (ICU). He was ventilated mechanically with catecholamine infusion. Two units of PRBC and 3 units of FFP were transfused. His state improved quickly and he was readmitted to the surgery department after 24 hours.

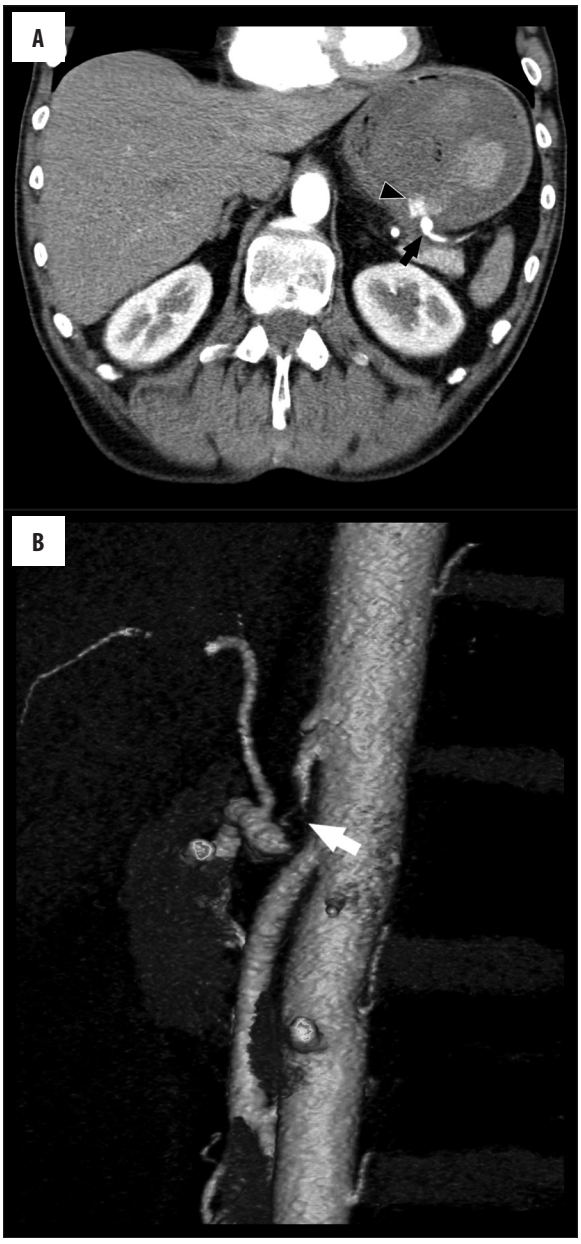
A follow-up ultrasound performed next week revealed a perigastric fluid collection 5.0×3.5 cm in size adjacent to the bleeding site. A follow-up CT performed two weeks after surgery showed regression of the pathological fluid collection to 2.8 cm in diameter (see Figure 2). The lesion was interpreted as a resolving hematoma being a typical complication of surgery. The ligated branch of the splenic artery with pseudoaneurysm was excluded from circulation. No signs of bleeding were observed. Interestingly, no splenic infarct was detected.

At that time, the patient was in a good condition, with a normal hemoglobin level, i.e. of 11.4 g/dL, without any symptoms of recurrent bleeding. He was discharged shortly thereafter.

Discussion

Pseudoaneurysm of the splenic artery is very uncommon. In a large series from the Mayo Clinic, 10 splenic artery pseudoaneurysms were found over 18 years [4]. By definition, in true aneurysms, the wall is composed of the intima, media, and adventitia; in comparison, a pseudoaneurysm wall consists of the intima and media only. Over 80% of splenic artery pseudoaneurysms are a consequence of pancreatitis or trauma. In the case of pancreatitis, pancreatic enzymes are thought to cause necrosis of the vascular wall and fragmentation of elastic tissues, leading to pseudoaneurysm formation. Additional mechanisms are related to pseudocysts found in 41% of patients with pseudoaneurysms [4]. A pseudocyst may induce a pseudoaneurysm not only through vascular erosion from enzymes within it, but also through direct compression or ischemia.

In the reported case, splenic artery pseudoaneurysm was diagnosed as a complication of a penetrating gastric ulcer. Presumably, gastric enzymes caused necrosis and weakening of a vessel wall, which resulted in pseudoaneurysm



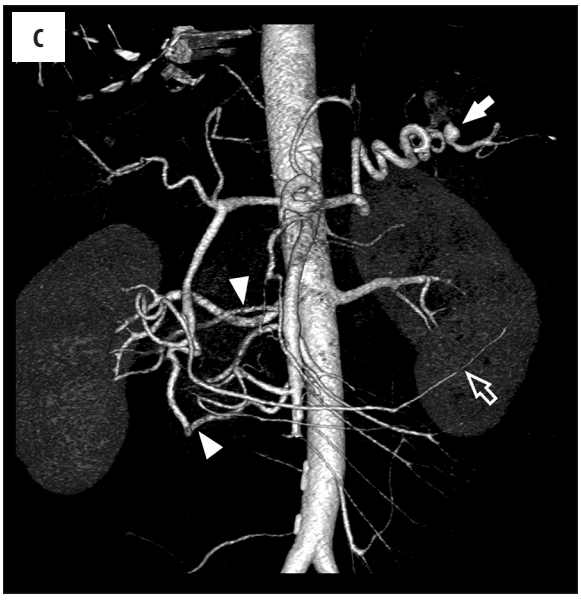


Figure 1. CT angiography: (**A**) A 5-mm oblique MPR shows the splenic artery pseudoaneurysm (arrow) on the posterior wall of the stomach with intragastric bleeding (arrowhead); (**B**) VRT reconstruction presents critical stenosis of the celiac artery (arrow); (**C**) VRT in coronal projection shows the ruptured pseudoaneurysm (arrow) with collaterals through the pancreaticoduodenal (arrowheads) and gastroepiploic arteries (empty arrow).

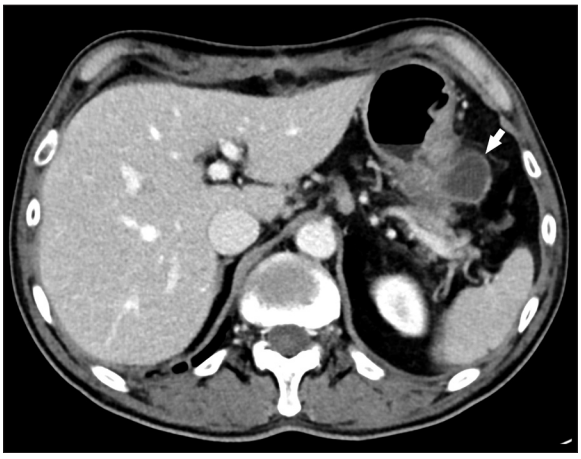


Figure 2. Follow-up CT in a 5-mm axial MPR shows epigastric pathological fluid collection (arrow) adjacent to the ligated vessel. The lesion was diagnosed as postoperative hematoma.

formation. The fistula was clearly evident intraoperatively, with the pseudoaneurysm plainly visible through the stomach wall. Peptic ulcer disease is a rare cause of splenic artery pseudoaneurysms; only four such cases have been reported.

Unlike true aneurysms, splenic artery pseudoaneurysms nearly always present with symptoms. Hemorrhage from a splenic artery pseudoaneurysm can lead to massive bleeding into the pancreatic duct, peritoneal cavity, retroperitoneal space or adjacent organs. The most common is bleeding into the pancreatic duct termed hemosuccus pancreaticus [4]. The majority of patients with bleeding (58%) are hemodynamically unstable at presentation.

The reported patient presented with symptoms of a massive GI hemorrhage. Bleeding stopped spontaneously with normalization of laboratory and hemodynamic parameters during the next 48 hours. However, the second episode of hemorrhage occurred two days later. Such a clinical course resembles the "double rupture" phenomenon, which was previously reported in true SAAs [7]. In true aneurysms, this phenomenon is explained by a tamponade of initial bleeding into the lesser sac, followed by a hemorrhage into the peritoneal cavity. In the described case, blood pressure in the splenic artery was initially decreased due to a critical stenosis of the celiac artery. This possibly allowed the hemorrhage to stop spontaneously. However, such an unusual clinical course was the reason for overlooking the source of bleeding on endoscopy and ultrasound performed

between the episodes of hemorrhage. That delayed the diagnosis and made the operative risk in an unstable patient extremely high.

According to the recent review, splenic artery pseudoaneurysm is the most commonly diagnosed by means of angiography (52%), followed by CT (36%). Interestingly, endoscopy and ultrasonography, although routinely performed in those cases, are rarely conclusive (0.7% and 3.3%, respectively) [4]. In the reported case, after a false negative result of endoscopy and ultrasound, the diagnosis was established with CT angiography. This supports a statement that angiographic techniques are the examinations of choice in diagnosing splenic artery pseudoaneurysms.

Aneurysms and pseudoaneurysms of the splenic artery require different management strategies. Most data suggest that treatment of a symptomatic SAA should be conducted in each case [4]. Moreover, several high-risk groups of asymptomatic patients should be considered for the repair, i.e. pregnant women or women of childbearing age, patients with a liver transplant or patients with cirrhosis and portal hypertension. However, no consensus has been reached regarding the intervention in asymptomatic patients with SAA. Recommendations are to treat asymptomatic aneurysms greater than 2 cm in patients with a reasonable operative risk and life expectancy of more than 2 years [4].

Because of the high risk of rupture and high mortality rate in case of splenic artery pseudoaneurysm rupture, the earliest possible intervention is deemed necessary. According to the review of 145 cases of splenic artery pseudoaneurysm, interventions (surgical and radiological) included embolization (37%), 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%). Ligation alone failed in 43% of cases in which it was attempted. Those patients underwent splenectomy, splenectomy and distal pancreatectomy or successful transcatheter embolization [4].

Surgical intervention carries mortality and morbidity risks of 1.3% and 9%, respectively [8]. Endovascular techniques using coils, detachable balloons, inert particles or gelatin sponge have reported success rates of 75–85% [8,9].

The presented patient was treated surgically with bipolar ligation of the bleeding branch of the splenic artery and suture of the gastric ulcer. Splenectomy was abandoned because the lesion was localized not in the trunk of the splenic artery but distally in one of its two main branches. Therefore, its ligation did not cut off the blood flow to the spleen completely as the other branch was still patent. Additionally, preoperative CT angiography revealed well-developed collaterals through short gastric and gastroepiploic arteries supplying the spleen distally to the ligated vessel. It was assumed that collateral supply would be sufficient to prevent postoperative splenic ischemia. The decision appeared to be right as no splenic infarction was detected in a follow-up CT. Endovascular embolization seemed to be an inadequate therapeutic option because it might have not represented definitive management in a patient with pseudoaneurysm with GI bleeding, as there was by definition a fistulous connection. Beside, there have been several reports of post-procedural coil migration in such situations [10]. In patients with splenic artery pseudoaneurysm involving adjacent organs surgery should be considered as definitive treatment to eradicate the underlying etiology and reduce further morbidity. Endovascular techniques may constitute a good option in cases without a coexisting pathology or in patients with a high operative risk.

Conclusions

The most important factor in identifying ruptured splenic artery pseudoaneurysm as the source of GI bleeding is considering the diagnosis. UGI hemorrhage from splenic artery pseudoaneurysm can have a relapsing course providing false negative results of endoscopy and ultrasound if performed between episodes of active bleeding. In such cases, immediate CT angiography is useful in establishing diagnosis and in application of proper therapy before possible recurrence.

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