

Spontaneous Pancreaticoduodenal Artery Pseudoaneurysm Rupture

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Background	A 50-year-old man presented with several months of abdominal pain that worsened over 3–4 days with intermittent nausea and vomiting. He reported a history of heavy alcohol consumption and had no recollection of any abdominal trauma in the recent past. Neither amylase nor lipase were elevated. Cross-sectional imaging revealed a ruptured visceral artery pseudoaneurysm with hematoma as well as peri-pancreatic edema consistent with pancreatitis in the pancreatic head. The patient underwent a visceral arteriogram and therapeutic embolization of a pseudoaneurysm localized to the inferior pancreaticoduodenal artery.
Summary	Visceral artery pseudoaneurysms typically develop secondary to other pathology, including iatrogenic trauma, pancreatitis, arteritis, or malignancies. The rate of rupture is 76.3%, with an incidence between 0.01 and 0.2% in adults. Visceral artery pseudoaneurysms can be diagnosed by ultrasound, CT, or visceral angiography, with sensitivities of 50%, 67% and 100%, respectively. Gastroduodenal artery/pancreaticoduodenal artery pseudoaneurysms in particular are rare variants of visceral artery pseudoaneurysms. When they occur more proximally in the gastroduodenal artery, they are more likely to be diagnosed before free rupture, but they carry a high mortality due to hemorrhage and hemodynamic instability when a free rupture occurs.
Conclusion	Visceral artery pseudoaneurysms are in the differential for abdominal pain. Although rare, acute free rupture of a visceral artery pseudoaneurysm can be lethal. Selected pseudoaneurysms may be managed nonoperatively, while definitive treatment requires angiography and arterial embolization.
Keywords	visceral artery pseudoaneurysm; gastroduodenal artery; pancreaticoduodenal artery; spontaneous rupture; hematoma; embolization

DISCLOSURE STATEMENT:

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Introduction

True aneurysms are disruptions in arterial walls that involve all three wall layers.¹ Pseudoaneurysms are formed from extravasated blood from the artery into a contained space outside the artery.¹ Unlike a true aneurysm, the wall of a pseudoaneurysm is formed from the surrounding tissues, not the wall layers of the artery from which the lesion arises.¹ Visceral artery aneurysms may arise secondary to collagen vascular disease, atherosclerosis, or hypertension.² In contrast, pseudoaneurysms are more commonly associated with trauma, pancreatitis, arteritis, or malignancy.^{2,3,4} The rate of rupture for aneurysms is 3.1%, while the rupture rate with visceral artery pseudoaneurysms is estimated at 76.3%. For pseudoaneurysms, size is not a reliable way to predict rupture.^{3,5} The prevalence of visceral artery pseudoaneurysms is between 0.01 and 0.2% and most are discovered incidentally.² The most commonly involved vessel for visceral artery pseudoaneurysms is the splenic artery, followed by the hepatic artery. Together, these sites comprise 60 to 70% of all visceral artery pseudoaneurysms.^{2,4} Other involved vessels in the order of more to less common include the superior mesenteric artery, celiac artery, gastropiploic artery, jejunal-ileal-colic arteries, pancreaticoduodenal artery, gastroduodenal artery and inferior mesenteric artery (Figure 1)⁴

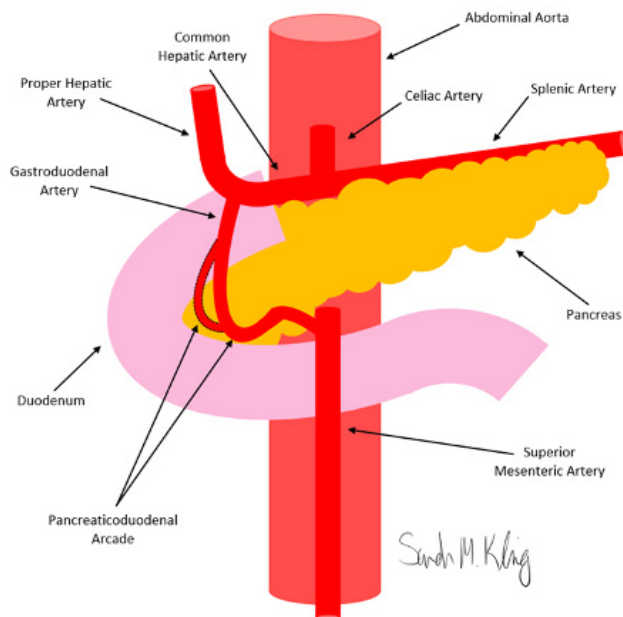


Figure 1. Figure 1. Representative anatomy of the gastroduodenal and pancreaticoduodenal arteries

Case Description

Here, we discuss a rare case of a ruptured pancreaticoduodenal artery pseudoaneurysm.

A 50-year-old man with a history of abundant alcohol intake presented with several months of intermittent abdominal pain that worsened over 3–4 days. This episode of epigastric pain came on one morning after drinking three vodka drinks the prior night. He noted intermittent abdominal pain roughly once per week over the last six months that he described as a feeling of vague peri-umbilical discomfort. These episodes always resolved on their own, and he never brought them to medical attention before now. He also suffered from intermittent nausea and vomiting for a week and lost 5–10 pounds over the prior few months. His last bowel movement occurred three days prior to presentation, but he continued to pass flatus. He denied back pain, syncope, or lightheadedness, and he had no prior abdominal surgeries. His medical history included hypertension, smoking 1.5 packs of cigarettes per day, and drinking 3–5 alcoholic beverages per day. He was not aware of any previous episodes of pancreatitis. His family history was notable for having a father treated recently for pancreatic cancer; a mother with breast cancer, anal cancer, and angiocarcinoma of the upper extremity; and a brother with testicular cancer.

On presentation, the patient's blood pressure was 137/104, and his hemoglobin was 11.7 g/dL. Amylase, lipase and lactate dehydrogenase were normal at 15 U/L, 15 U/L and 198 IU/L, respectively. Carcinoembryonic antigen (CEA) and cancer antigen 19-9 (CA 19-9) were normal at 6.9 ng/dL and 6.0 U/mL, respectively.

Imaging revealed peri-pancreatic edema consistent with pancreatitis in the pancreatic head. A triple phase CT of the abdomen and pelvis (Figure 2) demonstrated a hematoma involving the head of the pancreas, second and third parts of the duodenum. A pseudoaneurysm of the gastroduodenal artery was apparent. In light of this finding, he underwent a visceral arteriogram and embolization of the inferior pancreaticoduodenal artery and associated pseudoaneurysm, via a superior mesenteric artery approach. He recovered well and was discharged to home one day after the embolization. During the hospitalization he underwent alcohol cessation counseling.

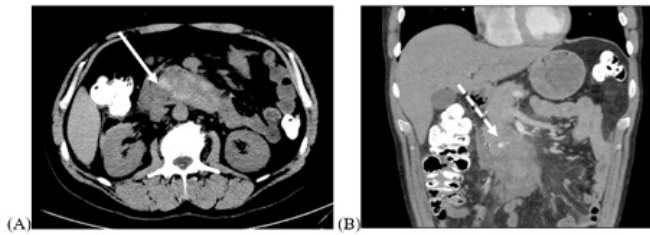


Figure 2. CT imaging pre-contrast (A) and with contrast in the arterial phase (B) showing evidence of a ruptured visceral artery pseudoaneurysm. (A) The axial section reveals a hyperdense area in the region of the inferior pancreaticoduodenal artery (solid arrow) in the absence of contrast, consistent with free blood. (B) The visceral artery pseudoaneurysm of the inferior pancreaticoduodenal artery (dashed arrow) is seen in the coronal section.

The patient subsequently returned to the hospital six days later with two days of recurring abdominal pain, abdominal distension, vomiting undigested material about 45 minutes after eating or drinking anything, constipation, weakness and light headedness. An upper gastrointestinal fluoroscopic study with contrast revealed slow passage of contrast with narrowing at the third part of the duodenum. The findings were consistent with a partial duodenal obstruction secondary to his resolving hematoma. He was managed without operative or endoscopic management, and he gradually was able to tolerate a full liquid diet over the next few days. At follow-up, he had complete resolution of his duodenal compression by upper gastrointestinal fluoroscopic study. One month after discharge, he was tolerating a regular diet.

Discussion

Visceral artery pseudoaneurysms present most commonly with abdominal pain, but they may also present with hypotension, back pain, jaundice, vomiting, gastric outlet obstruction, diarrhea, melena or hematochezia. The presentation is often dependent on the vessel location of the pseudoaneurysm and the route or location of a free rupture (if present). Hematomas within the lumen of the gastrointestinal tract, peritoneal cavity or retroperitoneum may account for such diverse symptomatology.^{5,6,7} Visceral artery pseudoaneurysms may be diagnosed by ultrasound, CT or visceral angiography with sensitivities of 50%, 67% and 100%, respectively.⁶ Since visceral artery pseudoaneurysms can undergo free rupture and result in hemodynamic instability, emergent treatment may be necessary.^{3,4} There is no correlation between pseudoaneurysm size and risk for free rupture.^{2,5}

Pseudoaneurysms of the gastroduodenal, or related pancreaticoduodenal arcade vessels, are extremely rare. They comprise roughly 5% of all visceral artery pseudoaneurysms.^{2,6} Most commonly, these abnormalities arise secondary to pancreatic inflammation related to chronic pancreatitis.^{2,6} Two-thirds of gastroduodenal artery pseudoaneurysms are diagnosed before free rupture occurs.⁶ The mortality rate of treated gastroduodenal artery pseudoaneurysms that have not undergone free rupture is 15%. However, the mortality rate reaches 50% when free rupture has occurred.^{4,6,8} Free rupture of pseudoaneurysms at these sites commonly occur into the bowel lumen (50% of cases) or peritoneal cavity.^{4,6,8} Consequently, they often present with gastrointestinal bleeding in the form of melena, hematochezia or hematemesis.^{sup>6,7,8} Gastroduodenal artery pseudoaneurysms presenting with rupture are emergencies in need of aggressive, early management due to the tendency to cause life-threatening hemorrhage, with associated hypotension and shock.^{6,7,8} When the pseudoaneurysm is localized more distally, in the pancreaticoduodenal artery arcade (as opposed to the gastroduodenal artery), the rate of rupture is 50–90%.² However, free rupture of the distal branches is usually retroperitoneal, contained and less commonly fatal.^{2,6}

In the literature, there have been four reported cases of gastroduodenal/pancreaticoduodenal artery pseudoaneurysm rupture, and additional retrospective studies mention ten others. The case reports included three men and one woman with ages ranging from 35 to 56 years. A 56-year-old woman presented with hypotension, had a pseudoaneurysm diagnosed by angiography, and was successfully treated with trans-arterial embolization.² A 54-year-old man presented with hypertension, later became hypotensive, had a pseudoaneurysm diagnosed by three-dimensional CT, underwent an unsuccessful embolization and expired as a result of the rupture.⁷ A 35-year-old man with chronic pancreatitis had a pseudoaneurysm diagnosed by CT. The pseudoaneurysm was treated with surgical ligation because the hospital did not have interventional endovascular capabilities.⁸ A 40-year-old man with acute pancreatitis and hemophilia A had a pseudoaneurysm diagnosed by CT and expired secondary to hematemesis before treatment.⁹ The pseudoaneurysms in the retrospective study were all visualized under angiography in patients with history of pancreatitis or prior pancreatic surgery.⁵ Out of the ten patients in the retrospective study, nine of them underwent successful embolization.⁵ The remaining patient's embolization failed due to intra-procedural rupture necessitating successful stent graft placement.⁵

The initial determination of treatment approach for patients with visceral artery pseudoaneurysms should be based largely on their hemodynamic status. Hemodynamically stable patients should undergo angiography with therapeutic embolization if possible. Surgical management may be considered for hemodynamically unstable patients, those with failed embolization, or when angiographic capabilities are not available.⁵

Embolization can be achieved with coils, particulate materials such as polyvinyl alcohol or gel foam, N-acetyl cyanoacrylate or a combination of these agents.^{5,11} Technical success is considered when perfusion of the pseudoaneurysm ceases no contrast extravasation is visualized.^{5,11} As an alternative to embolization, a stent graft may be deployed across the abnormal vessel to allow distal organ perfusion, while excluding the pseudoaneurysm from circulation.⁵ Stents cannot be used if there is not sufficient vessel length on either side of the pseudoaneurysm, if there is significant vessel tortuosity, or a correctly sized stent graft is not available.¹⁰

The technical success rates of endovascular intervention in two different studies ranged from 98–100%.^{5,10,8} Interestingly, there was no correlation between overall survival and the success of the embolization performed, although the sample sizes of the studies are small.⁵ In contrast, only low numbers of blood transfusion products needed prior to angiography are associated with better survival.⁵ Complications of embolization include end-organ infarction, post-embolization syndrome, re-bleeding and reperfusion of the pseudoaneurysm.^{10,11} Contraindications to endovascular intervention include the need to maintain patency of the feeding vessel or the presence of collaterals near or arising from the pseudoaneurysm. In these instances, a stent graft or surgical intervention may be needed.¹⁰

Surgical intervention includes pseudoaneurysm resection with revascularization, or pseudoaneurysm ligation with or without end organ resection.^{10,11} Postoperative morbidity and mortality is higher in patients treated with surgery, making endovascular approaches more appealing when possible.⁵

Conclusion

Herein, we describe a classic example of a retroperitoneal bleed from a pancreaticoduodenal artery pseudoaneurysm and associated free rupture. The patient was hemodynamically stable throughout. His pseudoaneurysm was most likely related to acute on chronic pancreatitis in a patient with a history of alcoholism. The abnormality was diagnosed by CT angiography, and treated successfully by trans-arterial embolization.

Trans-arterial embolization is the standard of care for treatment of visceral artery pseudoaneurysms. The technique is precise, successful, and has a low complication rate.^{4,5} Surgical intervention is a more invasive option, and generally reserved for patients with hemodynamic instability, where embolization is unsuccessful or not available.^{5,7,8}

Lessons Learned

Gastrooduodenal artery/pancreaticoduodenal artery pseudoaneurysms are rare forms of visceral artery pseudoaneurysms. When they occur more proximally in the gastroduodenal artery, they are more likely to be diagnosed before free rupture, but they carry high mortality due to hemorrhage and hemodynamic instability if they do undergo free rupture. Those of the more distal pancreaticoduodenal artery are more likely to be diagnosed after rupture but the bleeds are typically contained within the retroperitoneum and almost always survivable. This case highlights a visceral artery pseudoaneurysm, which should be considered in the differential diagnosis of abdominal pain and associated symptoms, especially in patients at risk for chronic pancreatitis. Diagnosis is effectively made by CT angiography, and successful management is achieved by trans-arterial embolization.

Abbreviations

Carcinoembryonic antigen (CEA) and cancer antigen 19-9 (CA 19-9) are tumor markers used in pancreatic cancer patients.

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