Hemosuccus Pancreaticus: A Case of Upper Gastrointestinal Bleeding Arising from a Pancreatic Pseudoaneurysm

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ABSTRACT

Pancreatic pseudoaneurysms are possible complications of chronic pancreatitis. These may present as hemosuccus pancreaticus, a rare cause of upper gastrointestinal tract bleeding where a pseudoaneurysm erodes into an adjacent pseudocyst or pancreatic duct, manifesting as bleeding through the pancreatic duct into the duodenal papilla. We report a case of a 32-year-old male with a history of chronic pancreatitis presenting as intermittent upper gastrointestinal tract bleeding secondary to hemosuccus pancreaticus from a pancreatic pseudoaneurysm. The patient underwent multiple sessions of endovascular embolization, which successfully controlled the bleeding despite some failed attempts; thus, a potentially morbid last-resort surgery was avoided.

Keywords: hemosuccus pancreaticus, pseudoaneurysm, chronic pancreatitis, upper gastrointestinal bleeding

INTRODUCTION

Chronic pancreatitis is a clinical syndrome characterized by progressive, long-standing inflammatory changes in the pancreas leading to structural damage and functional impairment. It has a wide spectrum of clinical manifestations, foremost of which are recurrent abdominal pain and pancreatic insufficiency, which may present as fat malabsorption and steatorrhea or as glucose intolerance and overt pancreatic diabetes. Other complications of chronic pancreatitis include pseudocyst formation, biliary and duodenal obstruction, pancreatic ascites and effusion, secondary pancreatic malignancy, and bleeding manifestations.[1] Pancreatic pseudoaneurysms are possible sequelae of chronic pancreatitis which may present as gastrointestinal bleeding. Management entails proper selection among available interventional and surgical options. We report the case of a pancreatic pseudoaneurysm showing as bleeding through the ampulla of Vater, discuss its development in relation to chronic pancreatitis, and review the different diagnostic and therapeutic modalities available.

CASE PRESENTATION

The patient is a 32-year-old male with a chief complaint of hematemesis. He had a laparotomy 22 years prior for a pancreatic pseudocyst and underwent endoscopic retrograde cholangiopancreatography (ERCP) with sphincterotomy 12 years ago, but other details were no longer available. The patient started experiencing recurrent episodes of cramping epigastric pain radiating to the back ten months before the

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consult. There were no associated fever, nausea, vomiting, jaundice, or bowel movement changes. Endoscopic ultrasound showed a pancreas with lobular borders and hyperechoic parenchymal foci, a dilated pancreatic duct measuring 2.9 mm with hyperechoic pancreatic duct walls, but without pancreatic duct calculi. Based on the constellation of symptoms and the presence of characteristic ultrasonographic findings, he was diagnosed with chronic pancreatitis and was started on medical management with relief of symptoms. One month before the consult, the patient started having intermittent episodes of hematemesis and melena. He was admitted to a local hospital for transfusion with 20 units of packed red blood cells and 8 units of fresh whole blood, then transferred to our institution for further management.

The patient was received with stable vital signs, pale palpebral conjunctivae, mild epigastric tenderness on direct palpation, and black tarry stools on the examining finger upon digital rectal examination. Laboratory tests showed normal hemoglobin and hematocrit levels post-transfusion, elevated total and direct bilirubin at 4.96 mg/dL and 3.77 mg/dL, alkaline phosphatase of 414 IU/L, and mildly elevated serum amylase and lipase levels at 132 and 731 U/L, respectively.

A tagged red blood cell scan showed abnormal focal tracer activity in the duodenal region on flow phase with tracer accumulation in the gastric area on the first hour of imaging with minimal tracer movement, suggestive of active bleeding in the duodenum and stomach. Upper gastrointestinal endoscopy showed pooling of blood within the gastric fundus and proximal body, normal gastric mucosa, and extrinsic compression at the junction of the first and second portions of the duodenum. On insertion of a duodenoscope, drainage of blood from the ampulla of Vater was visualized.

A computed tomography (CT) angiogram revealed an arterially-enhancing saccular outpouching at the region of the pancreatic head measuring 6.3 x 5.7 x 6.6 cm, with non-enhancing luminal hypodensities representing thrombus formation. The lesion appeared to arise from the gastroduodenal artery (GDA) and was causing secondary biliary and pancreatic ectasia (Figure 1). This was assessed as a GDA pseudoaneurysm, likely secondary to chronic pancreatitis. Mesenteric angiography was done, which showed a large ovoid area of contrast pooling representing the pseudoaneurysm arising from the proximal segment of the gastroduodenal artery (Figure 2). Superior mesenteric angiograms showed no additional supply to the pseudoaneurysm. The proximal segment of the GDA was occluded using coils and gelfoam particle embolization, with complete occlusion of the pseudoaneurysm post-embolization. The patient was discharged after embolization with no post-procedural complications.

Two weeks after discharge, the patient was readmitted for recurrence of the melena. A repeat CT angiogram showed a mild interval decrease in the size of the pseudoaneurysm at the peripancreatic region, with associated decrease in the biliary and pancreatic ectasia (Figure 3). Mesenteric angiography was repeated due to the persistence of the upper gastrointestinal bleeding. A small ovoid region of contrast pooling that was not present during the initial mesenteric angiography was seen arising from a duodenal branch of the superior mesenteric artery (SMA) (Figure 4). Subsequent contrast injections failed to demonstrate the said lesion, which was attributed to vasospasm of the SMA branch. This precluded embolization as a therapeutic

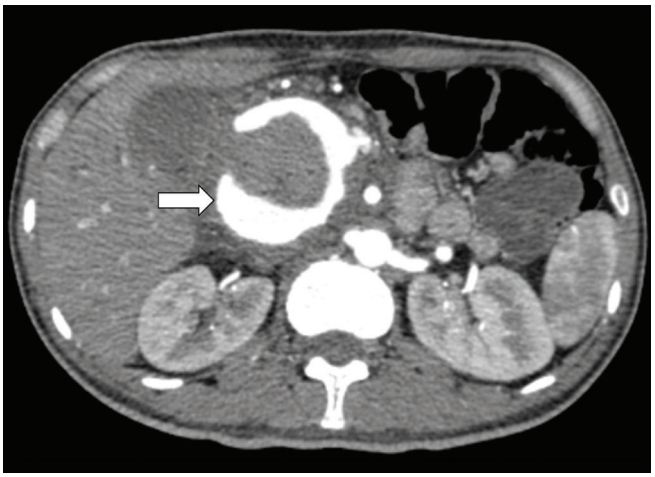

Figure 1. CT angiogram showing the pancreatic pseudoaneurysm *(white arrow)* at the peripancreatic region, with intraluminal hypodensities representing thrombus formation.

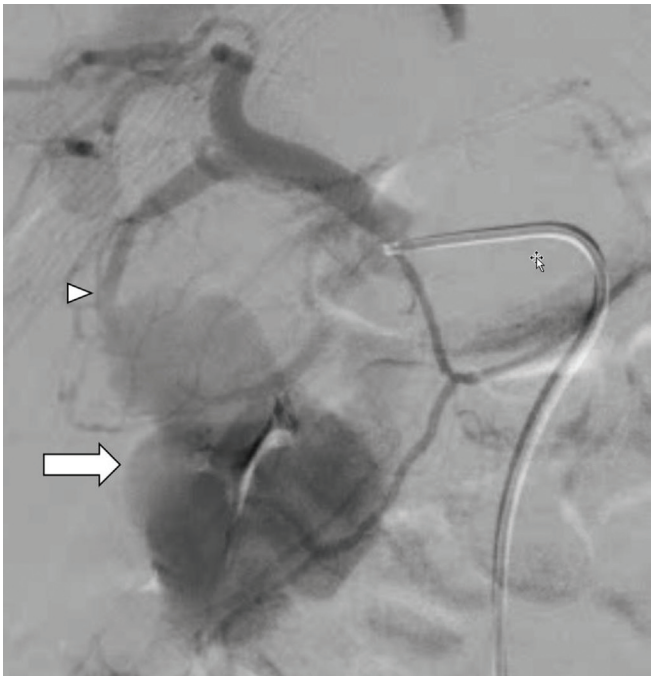

Figure 2. Pre-coiling angiogram showing the pancreatic pseudoaneurysm *(white arrow)* arising from the proximal segment of the gastroduodenal artery *(white arrowhead)*.

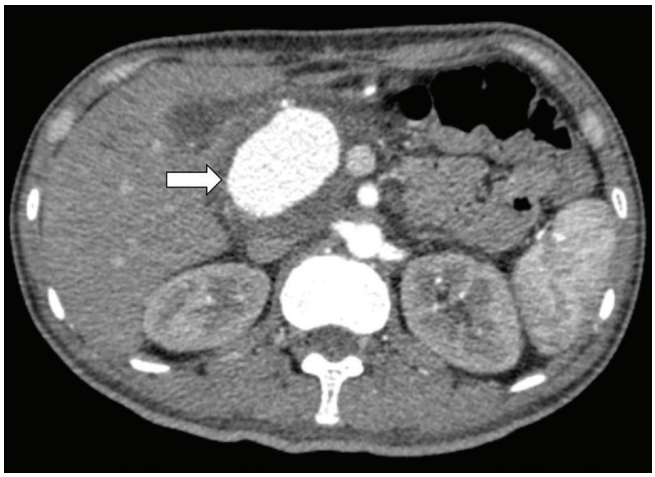

Figure 3. CT angiogram showing a mild decrease in size of the previously noted pancreatic pseudoaneurysm *(white arrow)*.

option. There was no opacification of the previously coiled GDA pseudoaneurysm.

Other therapeutic options such as surgery or intra-aneurysmal thrombin injection were considered since the first embolization failed to control the bleeding adequately. Repeat embolization could not be attempted during the second angiography. However, these options were deferred due to higher rates of complications, questionable efficacy, or lack of evidence. The multidisciplinary consensus was to reattempt embolization for the patient. A repeat mesenteric angiography was done, and the pseudoaneurysm from the distal duodenal branch of the SMA was successfully visualized and embolized using coils (Figure 5). The patient was discharged a few days after the procedure with no untoward events and has been seen on outpatient follow-up with no recurrence of bleeding.

DISCUSSION

Pseudoaneurysms are locally contained hematomas caused by damage to vessel walls. They are contained by surrounding structures and products of the coagulation cascade rather than portions of the vessel wall. Pancreatic pseudoaneurysms are known complications of acute or chronic pancreatitis, caused by arterial wall necrosis or exposure of the arteries to pancreatic elastases with subsequent degradation and extravasation.[2] These can occur in 3.5 to 10% of patients with pancreatitis.[2,3] Major vessels supplying the pancreas or within the peripancreatic region are at risk of developing pseudoaneurysms. Splenic artery involvement is the most common, followed by the gastroduodenal artery and pancreaticoduodenal artery, accounting for 40, 30, and 20% of reported cases, respectively. Pseudoaneurysms may rarely arise from the hepatic and gastric arteries, each comprising less than 5% of cases.[3]

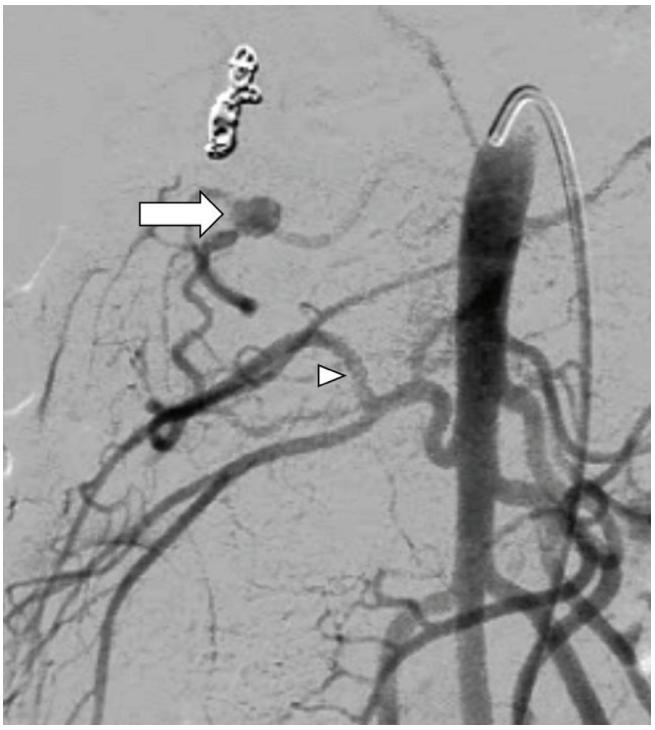

Figure 4. Angiogram showing a new smaller pancreatic pseudoaneurysm *(white arrow)* arising from a duodenal branch of the superior mesenteric artery *(white arrowhead)*. The coil deployed in the gastroduodenal artery is visualized with no opacification of the previous large pseudoaneurysm.

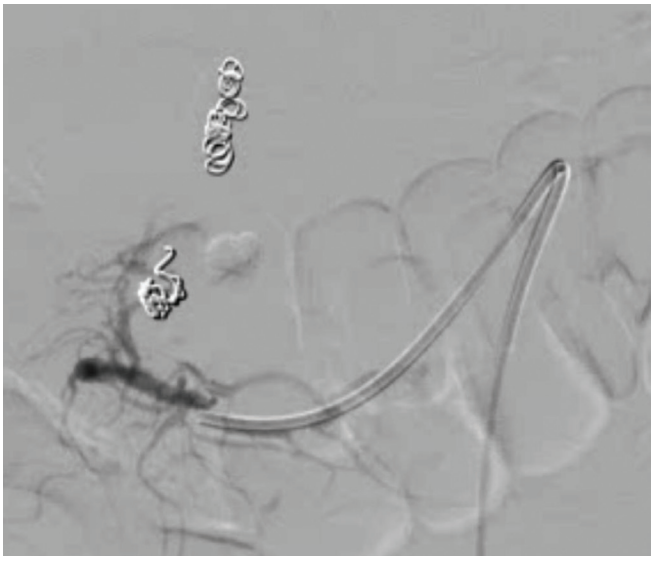

Figure 5. Post-coiling angiogram showing the successful coiling of the distal duodenal aneurysm, with no further opacification.

Pancreatic pseudoaneurysms may be complicated by rupture into adjacent organs or the peritoneal cavity in less than 10% of cases.[3] Bleeding may occur into pseudocysts with connections to the main pancreatic duct or directly into the ductal system, manifesting as bleeding from the ampulla.[4] The term hemosuccus pancreaticus (HP) was coined by Sandblom in 1970 to describe the phenomenon of bleeding from the pancreatic duct through the major duodenal papilla.[5] HP has also been referred to as pseudohemobilia, Wirsungorrhagia, or Santorinirrhagia in the literature. Hemosuccus pancreaticus caused by rupture of pancreatic pseudoaneurysms is a rare cause of upper gastrointestinal bleeding (UGIB), accounting for less than 1% of UGIB cases.[3] It is usually seen in males, with a mean age of incidence of 32 to 36 years.[6] HP occurs most commonly due to acute or chronic pancreatitis but can also be associated with vascular malformations, infections, pancreatic tumors, pancreatic divisum, pancreatic trauma, and iatrogenic causes such as endoscopic ultrasound-guided needle aspirations.[2] Pancreatitis is most strongly associated with cases of HP, and it has been suggested that HP should be considered in cases of chronic alcoholism with intermittent UGIB, even without evidence of chronic pancreatitis.[7] Clotting within the ampulla and pancreatic duct results in intermittent and repetitive bleeding, often not severe enough to cause hemodynamic instability despite the arterial origin. Patients may also present with biliary colic secondary to outflow obstruction and increased intraductal pressure from the intermittent clotting.[6]

Diagnosis of HP relies on identifying the bleeding source and delineation of the vascular anatomy. Endoscopy performed for cases of upper GI bleeding detects active bleeding via the ampulla in only 30% of patients with HP.[2] Ultrasonography and CT scan may show findings consistent with chronic pancreatitis, pseudocysts, and pseudoaneurysms. A sentinel clot, or clotted blood within the pancreatic duct on pre-contrast scans, is rarely seen, but its presence supports the theory of bleeding from the pancreas.[6] CT may also show opacification of a pseudocyst and the involved artery on contrast scans, and CT angiography can delineate the arterial anatomy. Direct angiography is still the gold standard for diagnostic purposes, identifying and localizing vascular lesions in the peripancreatic circulation with a 96% sensitivity rate.[2]

Untreated bleeding pancreatic pseudoaneurysms have a mortality of up to 90%.[4] Endovascular interventions are considered first-line treatment for pancreatic pseudoaneurysms.[4,8] Embolization with coils and other occlusive materials is the most commonly performed intervention. Other endovascular interventions such as balloon tamponade and stent grafting across the injured vessel are also options. This may also be combined with endoscopic drainage via transpapillary stenting to hasten the resolution of pseudocysts.[4] The suitability of embolization must be evaluated in cases where arterial supply might be compromised, and organ ischemia may be induced if there is inadequate collateral circulation. Placement of an intravascular stent across the aneurysmal vessel may be more appropriate in these cases, allowing for diversion of the bleeding while maintaining adequate perfusion.[4,7,8]

Endovascular interventions are less invasive and are considered appropriate for hemodynamically stable patients, in cases of diffuse bleeding or bleeding from the pancreatic head, after unsuccessful surgery or post-operative bleeding after surgical control of the pseudoaneurysm. Embolization for patients with transpapillary hemorrhage (such as hemobilia or hemosuccus) results in higher success rates versus for patients with primary upper gastrointestinal tract hemorrhage, with success rates for UGIB from pancreatic pseudoaneurysms ranging from 67 to 100%.[9] Overall mortality rates after embolization are at 4 to 19%, while rebleeding rates of around 30% have been reported.[2,4] Other complications related to endovascular interventions include rupture of the pseudoaneurysm during embolization, arterial perforation, intestinal necrosis, and aortic thrombosis. Factors associated with failure of embolization include multiorgan failure, anticoagulant use, corticosteroid use, pre-embolization vasopressor use, bleeding secondary to trauma or invasive procedures, longer time to angiography, previous surgery, and embolization with coils alone or coils with another agent.[10] Technical failures in embolization may result from difficult vascular anatomy, presence of arterial dissection, vasospasm, false-negative reading of an angiogram, multiple bleeding sites, or bleeding from a tumor.[11,12]

Surgical intervention for pancreatic pseudoaneurysms is indicated in cases of uncontrolled hemorrhage and hemodynamic instability, failure or unavailability of embolization, or for patients with other indications for operation such as pseudocysts, pancreatic abscesses or infected necrosis, gastric outlet obstruction, obstructive jaundice, or chronic pain.[2,9] Surgery may involve ligation of the bleeding vessel and excision of the pseudoaneurysm or pseudocyst, but a pancreaticoduodenectomy or distal pancreatectomy may be warranted in complicated cases. Identifying the offending vessel in a background of bleeding, inflammation, and necrosis caused by pancreatitis makes surgery technically difficult, with a high morbidity and mortality rate at 25 to 50%.[2,7] Mortality is higher in cases with the pseudoaneurysm located in the pancreatic head, whether arterial ligation or pancreaticoduodenectomy is performed.[6]

Given the difficulty and inherent risks of surgery for pancreatic pseudoaneurysms, other options have been explored should angioembolization fail. Intralesional thrombin injection with stimulation of thrombosis has been extensively studied in patients with femoral artery pseudoaneurysms after puncture for endovascular access, with successful bleeding control in almost 100% of cases.[13] This has led to the exploration of thrombin injection to manage visceral artery pseudoaneurysms. One early case report demonstrated the resolution of a pancreatic pseudoaneurysm at ten weeks follow-up after CT-guided percutaneous injection of 4000

U thrombin after the failure of embolization.[14] A series of 8 patients with visceral artery pseudoaneurysms (6 patients with a history of chronic alcoholic pancreatitis) managed with an endoscopic ultrasound-guided injection of 200 to 500 U thrombin showed obliteration of the pseudoaneurysm in 87.5% of cases at a median of 6 months follow-up.[15] Thrombin injection may be most appropriate in hemodynamically stable patients with a small pseudoaneurysm and a narrow vessel connection to minimize intravascular leakage of thrombin products. It may also be useful in patients with larger pseudoaneurysms or hemodynamically unstable patients as an adjunct to coil embolization.[14] Downstream embolization of coagulation products is a documented complication of thrombin injection, especially in cases with a short aneurysmal neck less than 3 mm or a wide origin from the involved vessel. Embolization may be avoided by injecting thrombin into the aneurysm sac away from the neck.[15] The use of thrombin injection in cases presenting as hemosuccus pancreaticus has not been investigated, and thrombin is not commercially available in the local setting.

CONCLUSION

Hemosuccus pancreaticus is a rare cause of upper gastrointestinal bleeding. It is strongly associated with cases of chronic pancreatitis with the formation of pancreatic pseudoaneurysms. It must be considered in patients with upper gastrointestinal bleeding with a history of pancreatitis or other pancreatic pathologies. Endovascular intervention is the preferred mode of treatment for pancreatic pseudoaneurysms, but alternative options must be considered in case of failure. It is prudent to reattempt endovascular intervention if no other interventions are viable, as surgery entails higher risk and potentially worse outcomes.

Statement of Authorship

Both authors contributed in the conceptualization of work, acquisition and analysis of data, drafting and revising and approved the final version submitted.

Author Disclosure

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