

Refractory hematemesis caused by haemoductal pancreatitis

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Abstract

We report a case of a 48-year-old female, who presented with refractory haemetemesis. Her oesophago-gastroduodenoscopy showed only a healing ulcer but profuse bleeding was seen from duodenum. In spite of a negative oesophago-gastroduodenoscopy she was bleeding profusely with haemodynamic decompensation. Doppler coeliac trunk showed a suprapancreatic cystic lesion with *yin-yang* pattern of blood flow confirming a pseudo aneurysm involving the superior and inferior pancreaticoduodenal arterial arcade using digital subtraction angiography. The conversion of a pancreatic pseudo cyst into a pseudo aneurysm is a potential lethal complication because, when rupture occurs, mortality rises up to 40%. She was diagnosed to have pancreatic pseudocyst, pseudoaneurysm and hemosuccus pancreaticus with wirsungorrhagia and was offered arterial embolization following which she improved. Patients with chronic calcificpancratitis (CCP) could remain silent and can present with normal amylase and lipase. Complications such as pseudocysts or pseudoaneurysms can be asymptomatic. The pancreas should be considered a possible site of hemorrhage in CCP in cases of refractory upper gastrointestinal haemorrhage. We highlight the importance of looking for causes other than bleeding duodenal/gastic ulcer/oesophageal varices in case of a refractory haemetemesis giving the patient option of a non-surgical modality of treatment and it's reduced risks. The effectiveness of embolisation for bleeding pseudoaneurysms is emphasized.

Introduction

When the differential diagnosis of upper gastrointestinal tract bleeding (UGIB) is discussed, the rupture of pancreatic pseudoaneurysms resulting in hemosuccus pancreaticus (hemoductal pancreatitis) and wirsungorrhagia are seldom mentioned. A high index of clinical suspicion is required to diagnosis this rare entity but is easy if Doppler/digital subtraction angiography (DSA) is done. We attempt to discuss the relevance of DSA and

the characteristic *Yin-Yang* sign, which may be decisive in contemplating non surgical options of treatment.

Case Report

A 48-year-old Asian female presented in the emergency department with recurrent episodes of hematemesis, melena, and abdominal pain for 2 weeks. The patient has been taking insulin since she was 8 years old for diabetes, with fair glucose control. On physical examination, she was conscious but anxious, with the following vital signs: pulse rate 140/min; blood pressure 100/70 mmHg; respiratory rate 19-20/min; and body temperature 99°F. The patient had severe pallor, jaundice, and epigastric tenderness. Laboratory investigations revealed a blood hemoglobin level of 7.7 g/dL, and normal total and differential counts. The peripheral blood smear showed normochromic normocytic anemia without abnormal cells. Bleeding and coagulation parameters were normal. Her blood sugar was 245 mg/dL, urea was 25 mg/dL, serum bilirubin was 7.5 mg/dL, and direct bilirubin was 2.9 mg/dL, with elevated serum alkaline phosphatase (665 U/L). Blood tests revealed her serum glutamic oxaloacetic transaminase and serum glutamic pyruvic transaminase were 64 IU and 34 IU, respectively. Her urinalysis and renal function tests were normal. Serum calcium and phosphorus, serum and urinary amylase, and serum and urinary lipase were normal. Esophagogastroduodenoscopy showed active bleeding duodenal ulcers (D1). Adrenaline (1:10000) was injected on four quadrants to stop the bleeding temporarily. Abdominal ultrasonograms showed mild intrahepatic biliary radical dilatation with dilated common bile duct with a cystic lesion of 3.6 cm x 3 cm in the suprapancreatic region. The gall bladder was grossly distended and contained heterogeneous deposits suggestive of hemobilia. The pancreas appeared atrophic, with calcified foci with a cystic structure adjacent to the cystic lesion, which is suggestive of chronic calcific pancreatitis with pseudocyst formation. Continued hematemesis prompted a repeat endoscopy on the third day, which showed a healing duodenal ulcer with no evidence of bleeding, but profuse oozing from the duodenum (D2). DSA of the celiac trunk was performed in the context of the healing ulcer and the oozing from the duodenum, which revealed a *yin-yang* (yin-yang sign) blood flow pattern diagnostic of pseudoaneurysm (Figure 1). It involves the superior and inferior pancreatic duodenal arterial arcade. Filling from both the superior mesenteric and the celiac artery with the extravasation of contrast into the duodenal lumen was observed (Figure 2).

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Key words: pseudoaneurysm, embolization, digital subtraction angiography, Hemobilia, haemoductal pancreatitis.

Contributions: CR, investigated and planned strategies of management, collected and compiled the data, wrote and formatted the manuscript; VG, TJ, assisted in data collection and compilation.

Received for publication: 23 August 2011.

Accepted for publication: 22 February 2012.

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Gastroenterology Insights 2012; 4:e6
doi:10.4081/gi.2012.e6

Embolization of the pseudoaneurysm was done. An Excel microcette was placed in the pancreatic duodenal arcade. A 3 mm x 8 mm standard helical coil and a 4 mm x 6 mm super soft helical coil were placed in the distal arterial lumen and the neck of the pseudoaneurysm. Approximately 2 mL of 25% glue was injected. The post procedure images (Figure 3) showed that the pseudoaneurysm and proximal arterial lumen was completely occluded. Final diagnosis: Chronic calcific pancreatitis (CCP), Hemosuccus pancreaticus (HP), pancreatic pseudocyst, and ruptured pseudoaneurysm with *wirsungorrhagia*.

The bleeding stopped and, on the second day, she took oral fluids. Within two days the melena cleared. She was treated with human insulin 30/70, 30 U in the morning and 24 U at bedtime and 40 mg pantoprazole twice daily before food intake. She remained apparently asymptomatic on discharge. She returned for follow-ups on the 3rd and 6th months, and the first year without any recurrence of symptoms.

Discussion

Dealing with UGIB is a challenge to physicians. Peptic ulcer disease (PUD) is responsible for 27-40% of all bleeding episodes.¹ Alcohol abuse, chronic renal failure, and/or no NSAID use increases the risk.² In approximately 80% of patients, bleeding from a peptic ulcer stops spontaneously.¹ Recurrent bleeding occurs in a minority of patients older than 60 years, the presence of shock upon admission, coagulopa-

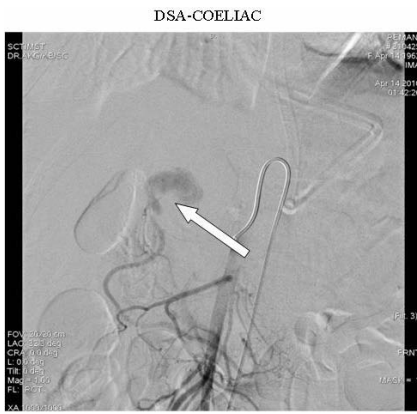


Figure 1. Showing yin-yang sign of pseudoaneurysm (thick white arrow).



Figure 2. Doppler/digital subtraction angiography image of pseudoaneurysm (thick white arrow) and other vessels labelled.



Figure 3. Post-embolisation image.

thy, active pulsatile bleeding, and presence of cardiovascular disease results in poor prognosis and a higher mortality rate.³ A study involving 10,428 cases of UGIB found that most patient deaths are not caused by bleeding. Therefore, the management of patients with UGIB should focus not only on hemostasis, but on lowering the risk of multiorgan failure and cardiopulmonary arrest.⁴ Risk factors such as recurrent bleeding, the need for endoscopic hemostasis or surgery, age older than 60 years, severe comorbidity, active bleeding, and hypotension are associated with increased mortality. Patients with hemorrhagic shock have a mortality rate of up to 30%.⁵ Pancreatic pseudoaneurysms are a very rare complication of CCP.⁶ Autodigestion of the peripancreatic artery or erosion of a pseudocyst into the artery and conversion of its cavity into a pseudoaneurysm result in HP (hemoductal pancreatitis),⁶ which is characterized by bleeding into the pancreatic duct, colicky pain, and jaundice.^{7,8} The term *hemosuccus* pancreaticus was coined by Sandbloom in 1970 upon discovery of three cases of bleeding from pancreatic duct.⁹ Until now, approximately 100 cases of HP have been reported.¹⁰ *Wirsungorrhagia* is gastrointestinal hemorrhage secondary to HP, wherein the blood from the pancreatic duct subsequently enters the intestines and the stomach. This is rare condition poses a diagnostic and therapeutic challenge.^{8,11} Doppler and DSA yields high-resolution images of the small arteries with the characteristic yin-yang sign.¹² Angiography has a 94.7% sensitivity and a 90% specificity. Embolization therapy is the treatment of choice regardless of etiology, location, or clinical presentation, which was successfully performed on the patient in the study.^{13,14}

Conclusions

Patients with CCP can present with normal amylase and lipase. Complications such as pseudocysts or pseudoaneurysms can be asymptomatic. The pancreas should be considered a possible site of hemorrhage in CCP and should be screened for ruptured pseudoaneurysms. The rarity of this complication is not a reason to underestimate the possibility of pseudoaneurysms. In refractory gastrointestinal bleeds, even if an ulcer is seen, when the GI bleeding continues, we suggest an early DSA to avoid unnecessary surgery and its associated risks.

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