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Hemosuccus pancreaticus: a rare cause of gastrointestinal bleeding

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Abstract

Hemorrhage from the pancreatic duct, referred to as hemosuccus pancreaticus or pseudohemobilia, is a rare cause of gastrointestinal (GI) bleeding. This potentially life-threatening complication of pancreatitis may pose a significant diagnostic and therapeutic dilemma, especially in patients who do not exhibit symptoms such as abdominal pain, jaundice, or GI bleeding. Here we describe a 55-year-old male with a known history of chronic calcifying pancreatitis, who presented with repeated episodes of melena associated with paroxysms of abdominal pain and frequent drop in hemoglobin requiring hospitalization. Initial endoscopic evaluation was negative. Endoscopy was repeated after an episode of melena which showed blood spurting from the ampulla. Further evaluation with abdominal CT scan, CT angiogram and conventional angiogram revealed no source of blood loss. Hence emergency surgery was done. There was evidence of splenic vein rupturing into the pancreatic duct.

Keywords Chronic calcifying pancreatitis, GI bleeding, hemosuccus pancreaticus

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Introduction

"Hemosuccus pancreaticus" is an unusual cause of severe upper gastrointestinal (GI) bleeding and results from rupture of splenic artery aneurysm into the pancreatic duct. Also known as pseudohemobilia and Wirsungorrhagia, it was first described in 1931 by Lower and Farrel who reported a primary splenic aneurysm rupture into the main pancreatic duct while the name hemosuccus pancreaticus was given by Sandblom in 1970. It is usually due to the rupture of a visceral aneurysm into the main pancreatic duct; splenic artery pseudoaneurysm associated with chronic pancreatitis represents the leading cause of this condition [1-3]. The association between pseudoaneurysm formation and pancreatitis is well established. Pseudoaneurysm occurs in 3.5% to 10% of cases of pancreatitis. However, rupture of a pseudoaneurysm is a relatively rare but life-threatening complication of chronic pancreatitis, occurring in 6-8% of patients with pseudocysts and accounting for less than 1% of cases of upper GI hemorrhage. If massive bleeding is untreated, the mortality rate is approximately 90%, whereas the mortality rate ranges from 25% to 37% in treated cases [4]. The intensity of bleeding ranges from intermittent occult bleeding up to massive acute bleeding causing death. The diagnosis is based on direct visualization of the hemorrhage through the main pancreatic duct at angiography. Given the intermittent nature of bleeding from pancreatic duct, delay in diagnosis may be fatal.

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Conflict of Interest: None

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Case report

A 55-year-old male diagnosed case of ethanol-related chronic pancreatitis presented with frequent episodes of melena associated with paroxysms of abdominal pain. He was given blood transfusions elsewhere for severe anemia. As patient had persistent GI bleeding with repeated drop in hemoglobin, he was referred for further evaluation and management. On clinical examination, he had profuse sweating, hypotension, tachycardia, severe pallor and mild tenderness in the epigastrium. His laboratory investigations revealed hemoglobin of 7.1 g% and hematocrit of 22%, mild renal failure and increased serum triglycerides. Contrast-enhanced computed tomography of the abdomen showed evidence of chronic calcifying pancreatitis (Fig. 1). Esophagogastroduodenoscopy with a standard scope and colonoscopy were normal. Repeat esophagogastroduodenoscopy with a standard scope after an episode of melena showed active bleeding from the ampulla (Fig. 2). Angiographic study could not localize the site of bleeding. Endoscopic ultrasound showed

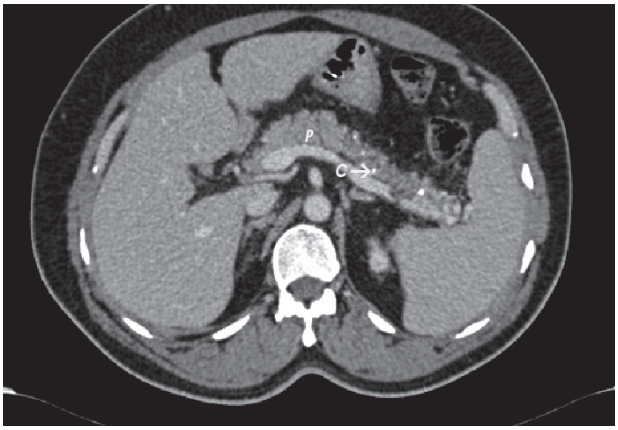

Figure 1 CT abdomen showing chronic calcifying pancreatitis
P, pancreas; C, calcification

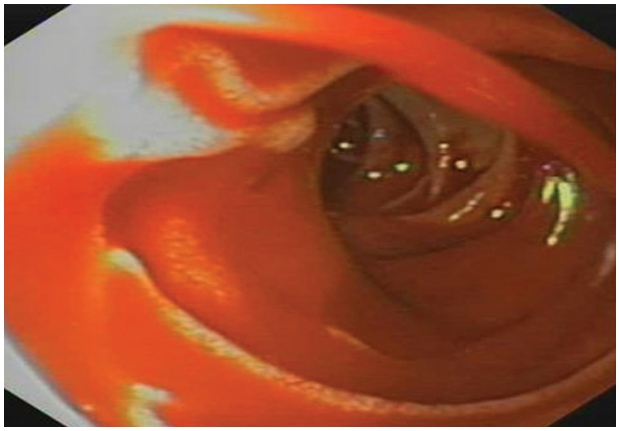

Figure 2 Esophagogastroduodenoscopy showing evidence of blood actively spurting from ampulla

chronic pancreatitis with a heterogeneous inflammatory lesion in the body of the pancreas (Fig. 3). As the bleeding source could not be localized and the patient had ongoing melena and drop in hemoglobin, emergency exploratory laparotomy was performed after initial hemodynamic stabilization. Pancreas was seen adherent to splenic vessels and hilum. There was evidence of splenic vein rupturing into the main pancreatic duct. Distal pancreatectomy with splenectomy was performed. The patient had an uneventful recovery postoperatively. Postoperative histology demonstrated chronic calcifying pancreatitis (Fig. 4A), lymphnodes with reactive hyperplasia and congested spleen (Fig. 4B). He was discharged in a hemodynamically stable condition and there was no further recurrence of GI bleeding after 6 months of follow up.

Discussion

Hemosuccus pancreaticus, defined as bleeding from the papilla of Vater via the pancreatic duct, is a rare and challenging cause of intermittent upper GI bleeding predominantly affecting men (male:female ratio 7:1) especially in relation to chronic alcohol intake. Causes of hemosuccus pancreaticus include acute and chronic pancreatitis, vascular malformation, pancreatic tumors (cystadenocarcinoma and osteoclastoma), pancreatic divisum, and iatrogenic or accidental trauma [5-7]. In patients with pancreatitis, pancreatic enzymes may cause necrosis of the peripancreatic vessels that cross tissue planes and boundaries, resulting in pseudoaneurysm formation. Chronic exposure of the arterial wall to digestive enzymes and the associated scarring and granulation on the pseudocyst create traction on the vessel, which may trigger GI hemorrhage. The arteries involved in GI hemorrhage in order of frequency include the splenic (40%), gastroduodenal (30%), pancreaticoduodenal (20%), gastric (5%), and hepatic arteries (2%) [8-10]. A pseudoaneurysm can rupture into the pseudocyst, GI tract, peritoneal cavity, or pancreatic parenchyma. Hemosuccus pancreaticus may be differentiated from hemorrhage due to pancreatic abscesses or stones eroding vessels in the wall of the GI tract. The hemorrhage is usually intermittent, repetitive and, most often, not severe enough to cause hemodynamic instability despite the usual arterial origin of bleeding. Hemosuccus pancreaticus is an entity diagnosed based on clinical, endoscopic and radiological findings, and a definitive diagnosis can be established only with angiography. However, hemorrhage from the papilla of Vater is rarely revealed with endoscopy and the fistula between the pancreatic duct and aneurysm of the peripancreatic vessels are seldom found with angiography and this explains why up to 52.9% of cases have not achieved a definitive diagnosis in some series [3]. There are two therapeutic options for this entity: surgery and angiographic embolization. Once the hemodynamic situation is under control, interventional radiographic methods are used for initial treatment, with immediate good results in 60-100% of cases. Coil embolization techniques provoke a thrombus in the aneurysm, but also obliterate the artery. Ischemia can develop in the tissue supplied by the artery if

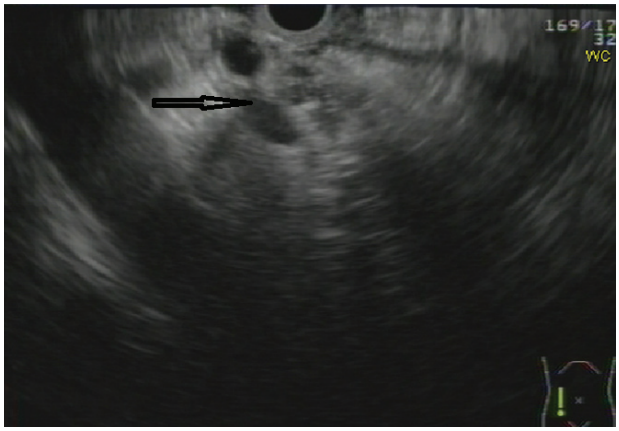

Figure 3 EUS showing evidence of chronic pancreatitis with inflammatory mass (arrow head showing inflammatory mass)

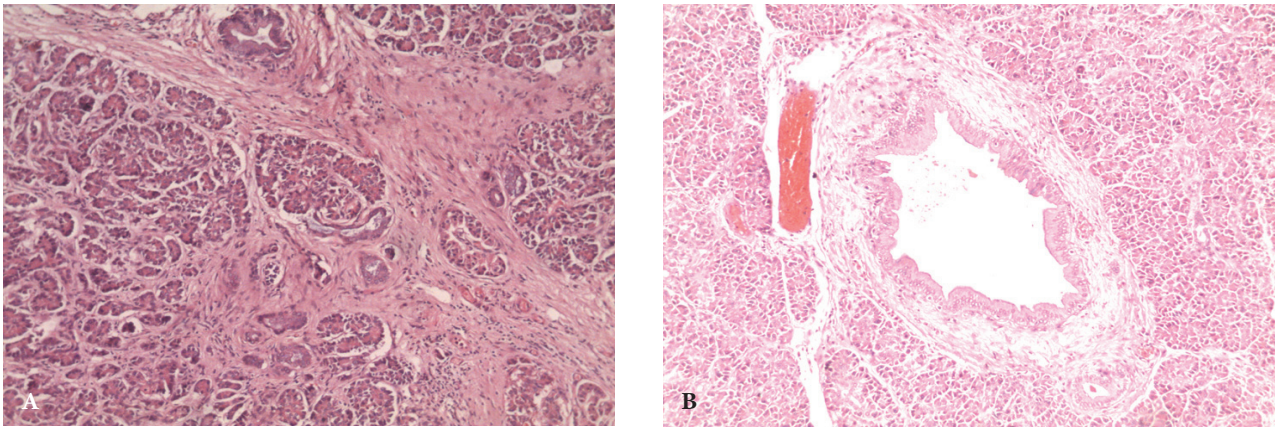

Figure 4 (A) Histopathologica examination showing evidence of chronic calcifying pancreatitis and (B) splenic congestion

the collateral circulation is not sufficient. Embolization of the celiac trunk, the common hepatic artery or the superior mesenteric artery is thus contraindicated. Surgical treatment is indicated in uncontrolled hemorrhage, persistent shock and when embolization is not feasible or when embolization fails (continued or recurrent bleeding), as well as in patients with other indications for operative intervention (pseudocyst, pancreatic abscess, gastric outlet obstruction, obstructive jaundice or incapacitating pain) who are otherwise appropriate surgical candidates [3]. Most surgical series have documented success rates of 70-85%, with mortality rates of 20-25% and rebleeding rates of 0-5%. A fistulous communication between the pancreatic duct and the splenic vein was found intraoperatively in this case, which is very rare and unusual.

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