CASE REPORT

HEMOSUCCUS PANCREATICUS

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Hemosuccus pancreaticus (HP) defined as bleeding into the pancreatic duct was first described in 1931 by Lower and Farell. HP also popularly known as wirsungorrhaghia and pseudohemobilia is a rare cause of gastrointestinal bleed. The unfamiliarity of this condition makes HP a diagnostic challenge. HP should be considered in patients with chronic pancreatitis presenting with acute gastrointestinal bleeding. The diagnosis is usually confirmed with a computerized tomography (CT) scan of the abdomen. A mesenteric angiogram with coil embolization can be performed to arrest the bleeding. The literature on this condition is restricted to case reports, case series and retrospective studies. We describe a case of HP in a patient with gastrointestinal bleeding and take this opportunity to review the literature outlining the diagnosis and management of HP.

Keywords: Hemosuccus pancreaticus; Gastrointestinal bleeding; Pancreatitis; Pseudoaneurysm

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INTRODUCTION

Hemosuccus pancreaticus (HP) defined as bleeding into the pancreatic duct was first described in 1931 by Lower and Farell. HP also popularly known as wirsungorrhaghia and pseudohemobilia is a rare cause of gastrointestinal bleed. The unfamiliarity of this condition makes HP a diagnostic challenge. HP should be considered in patients with chronic pancreatitis presenting with acute gastrointestinal bleeding. The diagnosis is usually confirmed with a computerized tomography (CT) scan of the abdomen. A mesenteric angiogram with coil embolization can be performed to arrest the bleeding. The literature on this condition is restricted to case reports, case series and retrospective studies. We describe a case of HP in a patient with gastrointestinal bleeding and take this opportunity to review the literature outlining the diagnosis and management of HP.

CASE

A 51-year-old male with a history significant for chronic pancreatitis, alcohol abuse and iron deficiency anaemia was admitted to the hospital with a history of coffee ground emesis and intermittent abdominal pain located in the right upper quadrant and epigastric region.

A year ago, he was admitted for severe acute blood loss anaemia and a workup revealed an unremarkable upper and lower gastrointestinal endoscopy (gross and microscopic) and a negative nuclear medicine tagged RBC scan. He appeared pale and tachycardic with a positive orthostatic blood pressure change. The abdomen was not distended; however, there was guarding and tenderness on palpation of mid-epigastric area. No stigma of chronic liver disease was noted. Rectal examination showed dark brown stool, which was positive for occult blood. Examination of other organ systems was unremarkable. Pertinent laboratory values indicated haemoglobin of 7.0 g/dL, white cell count elevated to 20.0x1000/uL and a lactate of 1.7 mMol/L. His urea was disproportionately elevated at 62 mg/dL, and creatinine was 2.0 mg/dL (baseline creatinine of 0.5 mg/dL) and an elevated anion gap of 28. His liver function tests were normal. A CT scan of the abdomen showed acute on chronic pancreatitis with increased density of blood within the second portion of the duodenum (Figure-1). On the Magnetic resonance imaging (MRI) abdomen a large pseudoaneurysm arising from the distal aspect of the gastroduodenal artery (GDA) in close relation to the inferior aspect of the pancreatic head and a smaller pseudoaneurysm arising from the inferior pancreaticoduodenal artery (Figure-2). Interventional radiology guided Micro coil embolization of the pseudoaneurysm in the gastroduodenal artery was successfully performed. The patient was eventually discharged home after a prolonged hospital stay.

Three months later, he presented with hematemesis, and his haemoglobin on presentation was 4.1g/dL. Upper endoscopy performed showed bright red blood in the duodenum and side viewing of the endoscopy showed bleeding originating from the ampulla of Vater. Repeat SMA angiogram (Figure-3) showed no active bleeding. Prophylactic embolization of the mid splenic artery was

performed. Despite these interventions, he had ongoing intermittent bleeding requiring multiple transfusions. His complicated abdominal vasculature and occluded celiac artery made him a poor candidate for the Whipple's procedure or any surgical interventions. A repeat MRI of the abdomen showed signs of Cholangitis, splenic infarction, and abscess. (Figure-4). The patient subsequently developed sepsis secondary to acute cholangitis with Enterococcus faecalis bacteraemia. Eventually, he succumbed to a cardiac arrest and passed away.

DISCUSSION

Our case reflects the typical presentation of HP, with intermittent abdominal bleeding in a patient with a history of chronic pancreatitis. Initial diagnostic work up failed to identify the cause of the bleeding. This case highlights the failure of IR guided coiling and outlines the complications associated with the procedure. Hemosuccus pancreaticus (HP), characterized by bleeding from the ampulla of Vater via the pancreatic duct, is an extremely rare cause of gastrointestinal bleeding, occurring in about 1 in 1500 patients presenting with upper gastrointestinal bleeding.[1] Due to the rarity of the condition, the literature on HP is mainly from retrospective studies, case reports, and case series. HP most commonly occurs as a consequence of pseudoaneurysm formation due to chronic pancreatitis.[2] The other common causes of HP are listed in table-1.

Rammohan and colleagues in the largest retrospective on HP describe the most common presentation to be melena, symptomatic anaemia, and abdominal pain.[1] Intermittent epigastric pain and bleeding, one of the characteristic signs of HP, is due to transient pancreatic duct obstruction and with relief of pain within 48 hours following passage of the blood clots and bleeding. The other symptoms and signs include nausea, vomiting, weight loss, and icterus.[3] During a flare-up of pancreatitis, the proteolytic enzymes can cause autodigestion of a vessel wall resulting in a pseudoaneurysm. These pseudoaneurysms can erode into the pancreatic duct resulting in HP. HP in patients with chronic pancreatitis with pseudocysts can be secondary to erosion of the pseudocyst into peri-pancreatic artery or formation of an arterial aneurysm that may erode into the pancreatic duct. Direct pressure erosion from the pseudocyst can also result in HP.[2] Pseudoaneurysms most commonly involve the Splenic artery (60–65%) followed by the GDA, Pancreaticoduodenal, Hepatic, left gastric and superior mesenteric artery.[2] Both GDA and Pancreaticoduodenal artery aneurysms are seen more frequently in the presence of celiac artery stenosis or occlusion.[4]

Due to intermittent nature of symptoms the diagnosis remains a challenge despite the availability of various diagnostic modalities (Table-2). Bleeding through the ampulla of Vater and/or the presence of blood in the second portion of the duodenum without an obvious source during endoscopy is suggestive of possible HP. Upper GI endoscopy is normal in nearly 50% of the patients.[1] Endoscopic retrograde cholangiopancreatography (ERCP) may show filling defects within the pancreatic duct, suggestive of blood clots within the pancreatic lumen or due to compression from a pseudo aneurysm.[5] Endoscopic ultrasound (EUS) has been useful in demonstrating the presence of a fistula between an aneurysm and pancreatic duct.[6] Contrast-enhanced EUS has been shown to be useful when angiogram has failed to delineate the feeding artery from the pseudo aneurysm.[7] CT scans, and MRI can be helpful in determining local anatomy, distinguishing pseudocysts, and identifying pseudoaneurysms. The "sentinel clot sign" is characteristic of HP and is clotted blood in the pancreatic duct on CT scans.[8] Few case reports support the utility of MRI as an alternative to CT as a diagnostic modality.[8,9] In addition to being radiation free, a properly performed MRI of the abdomen provides several diagnostic advantages. Using pre-contrast, fat-saturated T1w Gradient echo imaging, MRI offers a powerful imaging tool for the detection of small quantities of blood products within the pancreatic duct and the C-loop of the duodenum. Furthermore, MRI is the imaging modality of choice for evaluating the ampulla, distinguishing pseudocysts from abscesses, and evaluating for extent of pancreatic necrosis.[10–12] The utility of radionuclide red blood cell scintiscan is limited due to the intermittent nature of symptoms.[5] Angiography is routinely performed to diagnose HP. Despite its high sensitivity, the intermittent nature of symptoms limits its utility in localizing the site of bleeding.[2]

During angiography interventions include embolization, percutaneous thrombin injection, balloon obstruction, and covered stent grafting.[13] Interventions in the form of coil embolization, balloon tamponade, and placement of prosthetic material, can either be curative or a temporary measure before surgery in advanced cases. Review of the literature suggests that surgery was the mainstay treatment for HP in the 1990s. However, with the increasing experience in angiography, the success rate is as high as 75–100% with relatively low morbidity (14–25%) and mortality (0–33%).[1,14] Ischemia and infarct can develop if the collateral circulation is insufficient. However, this can be prevented by the use of stents; especially, non-coated metallic stents that favour the formation of a new arterial intima.[15]

Surgical may be warranted for uncontrolled haemorrhage, hemodynamic instability, or in situations where embolization has failed to resolve the bleed. In certain cases when we have other

concomitant operative indications such as a pancreatic pseudocyst, pancreatic abscess, gastric outlet obstruction, malignancies, etc., surgery might be the best therapeutic approach.[1,3,16] Intraoperatively- intracystic ligation, external ligation of the feeding vessels can arrest the bleeding. In certain situations, more aggressive surgeries, namely distal pancreatectomy or pancreaticoduodenectomy may be needed to prevent recurrence of bleeding. Proximal and distal arterial ligation is not recommended in patients with GDA and PDA aneurysms due to high recurrence rate for bleeding.[2]

Figure1: Coronal noncontrast CT. There are coarse calcifications in the enlarged and edematous pancreas, consistent for acute on chronic pancreatitis. Increased density or blood (short arrow) is present within the wall of the second part of the duodenum. This results in obstruction with a markedly distended stomach (large arrow).

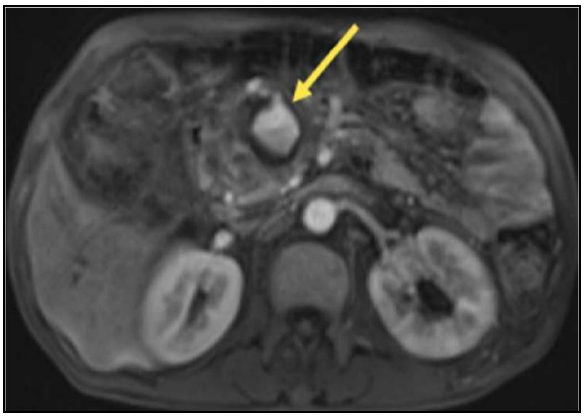

Figure-2: (A) MRI image (T1w 20 second arterial phase). There is a large pseudoaneurysm (arrow) originating from the gastroduodenal artery. This pseudoaneurysm exerts a mass effect upon the common bile duct and the duodenum.

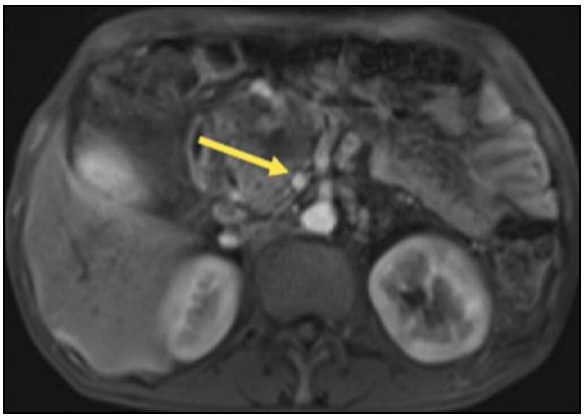

Figure-2: (B) MRI image (T1w 20 second arterial phase). A small pseudoaneurysm (arrow) arises from the inferior pancreaticoduodenal artery.

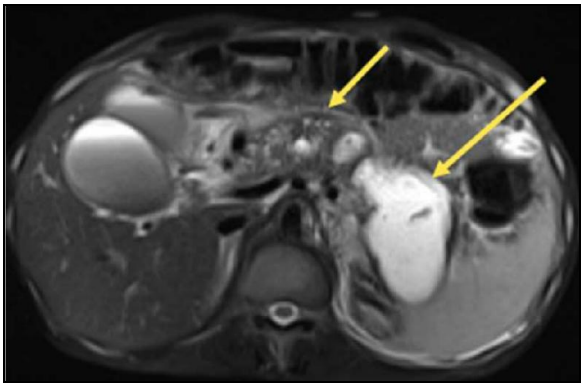

Figure-2: (C) MRI image (T2w fat saturated). A sizeable pseudocyst (large arrow) is identified at the tail of the pancreas. There is an acute inflammatory signal (small arrow) at the head and neck of the pancreas. Pronounced reactive inflammation extends into the porta hepatis and pancreaticoduodenal groove.

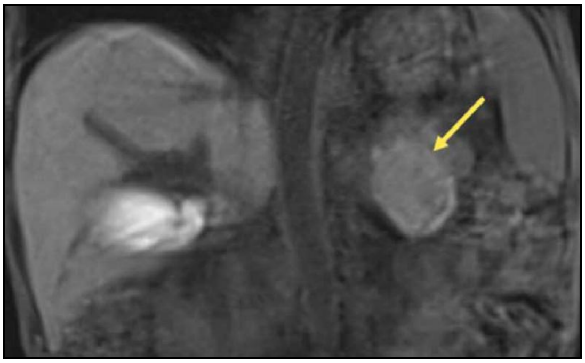

Figure-2: (D) MRI image (Coronal T1w precontrast). Extensive debris and blood products (small arrow) dependently layer within the pseudocyst.

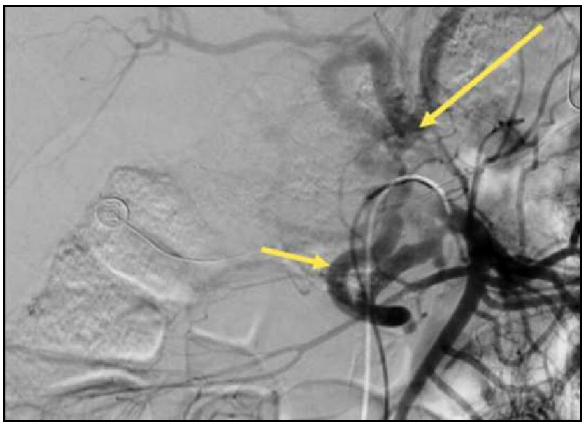

Figure-3: (A) Digital Angiogram. The superior mesenteric artery is cannulated with a catheter. The inferior pancreaticoduodenal artery (small arrow) is enlarged and supplies collateral blood supply to the celiac artery (large arrow) via the gastroduodenal artery. Contrast opacification of the hepatic artery and splenic artery is demonstrated. The celiac artery is occluded at its origin from the aorta.

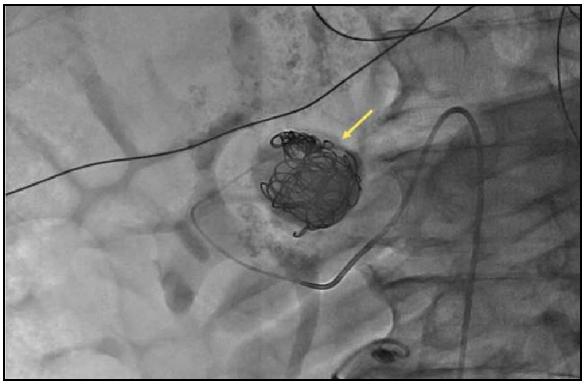

Figure-3: (B) Digital Angiogram. Embolization coils have been placed into the large pseudoaneurysm which arises from the gastroduodenal artery.

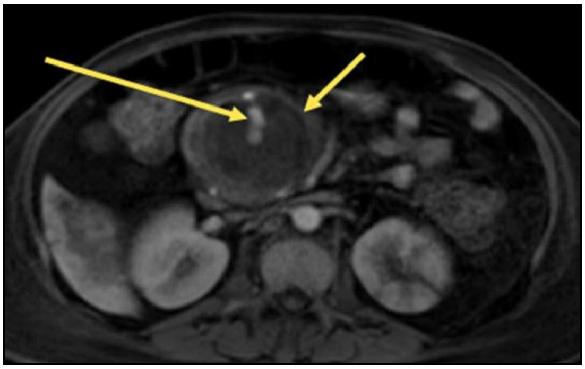

Figure-4: (A) MRI image (T1w 20 second arterial phase). The large pseudoaneurysm (small arrow) arising from the gastroduodenal artery is near completely thrombosed by the previously placed embolization coils. There is a small blush of active bleed (large arrow) extending into the thrombosed pseudoaneurysm.

Figure-4: (B) MRI image (Coronal T1w 5 min delayed phase). The common bile duct and intrahepatic biliary tree displays pronounced wall thickening (small arrows), consistent with cholangitis. Secondary to the longstanding obstruction of the distal common bile duct at the level of the pancreatic head, there is intrahepatic and extrahepatic biliary dilatation with several peripheral fluid collections, consistent with bilomas (large arrows).

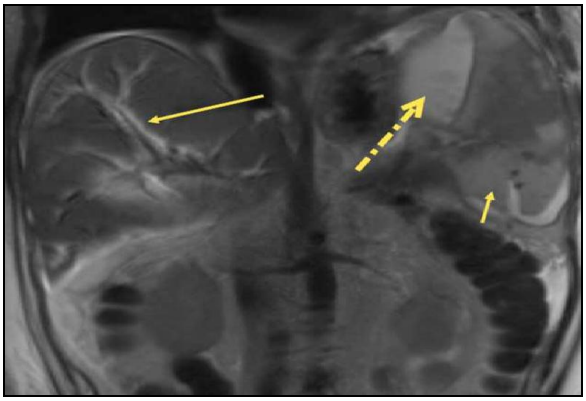

Figure-4: (C) MRI image (Coronal T2w). Periportal edema insinuates along the biliary tree (long arrow). There is infarction of the inferior spleen (small arrow), illustrated by the high T2 signal. Along the gastrosplenic ligament, a rim-enhancing fluid collection is demonstrated, consistent with an early abscess (dashed arrow). (

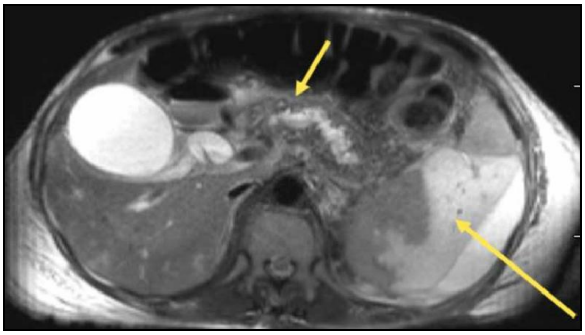

Figure-4: (D) MRI image (T2w fat saturated). The pancreatic duct is markedly distended and irregular (small arrow). There is pronounced inflammatory signal within and surrounding the pancreas. The inferior portion of the spleen is infarcted (large arrow).

Table-1: Common causes of Hemosuccus Pancreaticus

Diagnosis of Hemosuccus Pancreaticus
1. Upper Gastrointestinal Endoscopy – normal in 50% of patients
2. Angiography – highest sensitivity; Diagnostic and therapeutic.
3. Ultrasound
4. Abdominal CT and MRI – The first test of choice; Helpful in determining the local anatomy, identifying pseudocyst or an aneurysm
5. Radionuclide Testing

Table-2: Diagnostic modalities for Hemosuccus Pancreaticus

Causes of Hemosuccus Pancreaticus
1. Pseudoaneurysm of the peripancreatic arteries due to acute or chronic pancreatitis
2. Trauma
3. Rupture of a true aneurysm
4. Pancreatic tumours
5. Arteriovenous malformations

Table 3: Key points

Key points
1. Recognize the common clinical presentation of this uncommon condition
2. Systematic approach to diagnosis and management
3. Coordinate early care with Gastroenterology, Intervention radiology, and General Surgery
4. Anticipate complications related to various interventions

CONCLUSION

Our patient's presentation was unique and challenging. His high-grade occlusion of the celiac artery resulted in collateral blood supply to the liver through the superior mesenteric artery and gastroduodenal artery. Any surgical intervention including Whipple's procedure, could not be safely completed with his complex abdominal vasculature. HP is a rare and uncommon cause for GI bleeding. If not diagnosed in a timely fashion, HP carries high morbidity and mortality.

Health care providers in the emergency department, hospital medicine should familiarize with HP and its presentation. When HP is suspected, the first diagnostic step should be cross-sectional imaging of the abdomen followed by IR consultation for mesenteric angiography. If bleeding continues, consultation with the surgical services would be warranted.

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