

CASE REPORT

Hemosuccus Pancreaticus Due to a Noninflammatory Pancreatic Pseudotumor

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ABSTRACT

Context Hemosuccus pancreaticus is a rare source of gastrointestinal bleeding, the most frequent cause of which is pancreatitis, followed by tumors, but nearly all these tumors are true neoplasms, and not pseudotumors. Furthermore, nearly all pseudotumors of the pancreas and retroperitoneum are inflammatory. **Case Report** We present a case of hemosuccus pancreaticus associated with a nonneoplastic noninflammatory pseudotumor of the pancreas. **Conclusions** Pancreatic pseudotumors are not always inflammatory and should be considered in the differential diagnosis of gastrointestinal bleeding associated with hemosuccus pancreaticus.

INTRODUCTION

Hemosuccus pancreaticus (HP) is bleeding from the ampulla of Vater via the pancreatic duct. Also known as pseudo-hemobilia and Wirsungorrhagia, the condition was initially described in 1931 by Lower and Farrell [1], while the name hemosuccus pancreaticus was coined by Sandblom in 1970 [2], in his report of three cases of splenic-artery aneurysms which ruptured into the pancreatic duct. Pancreatic pseudotumor is a mass-forming lesion of the pancreas that is generally nonneoplastic, and robustly inflammatory. They are well known for mimicking pancreatic neoplasms, and indeed are the primary finding following pancreatectomy performed with a preoperative diagnosis of pancreatic cancer [3].

CASE REPORT

A 47-year-old African American male was admitted to the hospital for left chest wall abscess and diabetic ketoacidosis. Past medical history included diabetes mellitus type 1, essential hypertension, paroxysmal atrial fibrillation, ischemic stroke with minimal residual right sided paralysis, and chronic kidney disease. During the appropriate treatment of his admitting complaints, and management of his comorbidities, which included anticoagulation for the history of paroxysmal atrial fibrillation with CHADS score of 4, his hemoglobin dropped precipitously from 10 g/dL to 8.5 g/dL, then stabilized, then again dropped to 6.7 g/dL. Evaluation included esophagogastroduodenoscopy, which revealed fresh

blood from second portion of duodenum, emanating from the ampulla (Figure 1). Contrast-enhanced MRI (Figure 2) showed a solid lesion within the pancreatic tail, measuring 1.8 cm x 1.5 cm, hyperenhancing and consistent with a pancreatic neuroendocrine tumor (PNET); there was no liver lesion identified. Endoscopic ultrasound (EUS) was concordant regarding size, shape, and location of the pancreatic lesion, and furthermore showed no stones, masses, or dilatation of the common bile duct (Figure 3). Biopsy of the pancreatic tumor was not performed due to concern for bleeding.

The patient underwent a totally laparoscopic, spleen- and vessel-preserving distal (left) pancreatectomy. Intraoperative and gross pathological findings (Figure 4) were concordant with preoperative imaging corresponding to 1.8-cm lobular mass at the tip of the pancreatic tail. The patient tolerated the procedure well and was discharged on fourth post-operative day without complications. At short-term follow-up (2 months) he has had no further episodes of bleeding.

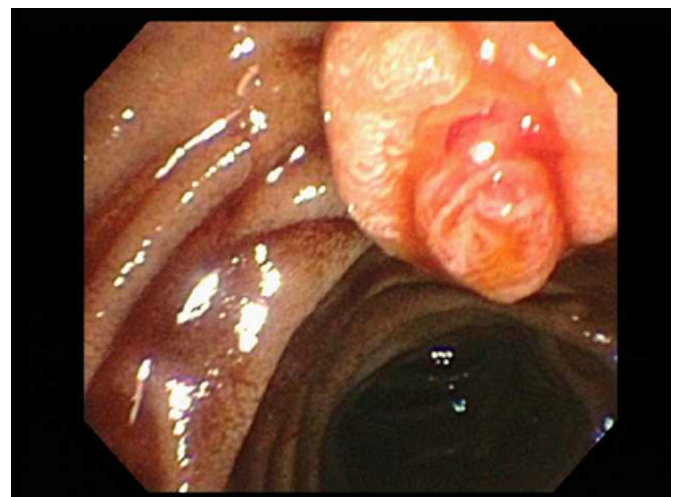


Figure 1. View at EGD, with gross blood emanating from major papilla.

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Key words Gastrointestinal Hemorrhage; Pancreas; Pancreatectomy; Pancreatic Ducts; Pancreatic Neoplasms

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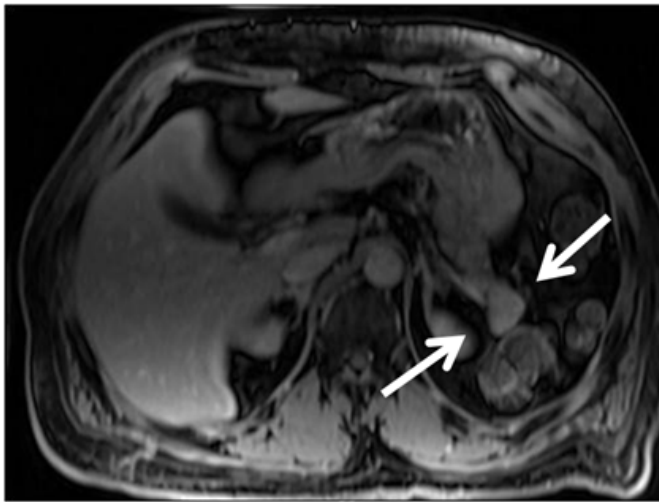


Figure 2. Early-arterial-enhanced nodule within pancreatic tail. Postcontrast VIBE image showing the enhancing nodule (arrows), with signal characteristics distinct from the spleen (not shown), precluding intrapancreatic splenule.

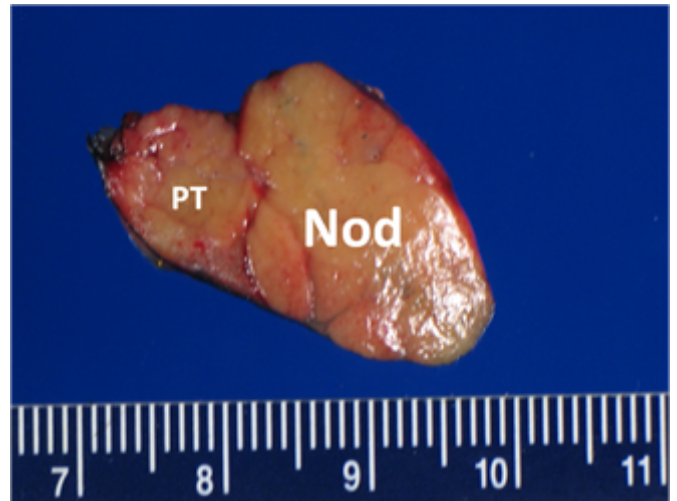


Figure 4. The resected, bisected specimen, showing the nodule (Nod); and the pancreatic-tail (PT) margin with staple line.



Figure 3. Endoscopic ultrasound showing a nodule (Nod) within the pancreatic tail, concordant with MRI, adjacent to the splenic artery (SA).

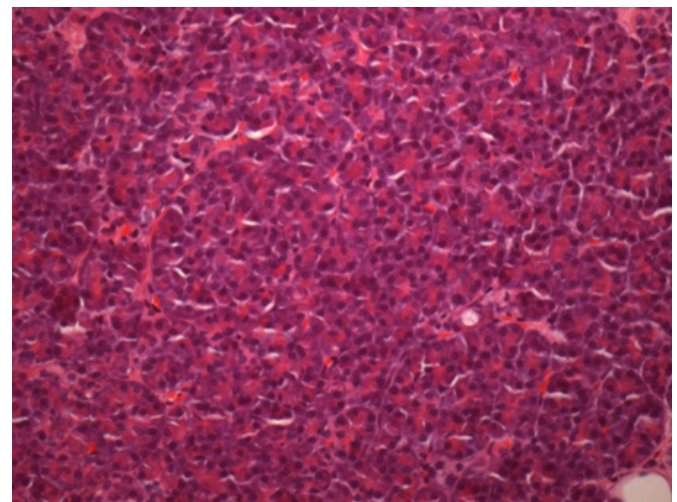


Figure 5. Photomicrograph of pseudonodule, stained with hematoxylin and eosin, showing noninflamed pancreatic parenchyma, with no neoplasm identified.

DISCUSSION

There are many potential causes of HP including pancreatitis (acute and chronic), neoplasms, vascular disease, trauma, infection, iatrogenic, and congenital abnormalities. Pancreatitis is the most common cause and is usually a result of pseudoaneurysm secondary to inflammation with eventual communication with the pancreatic duct [4, 5]. The most commonly involved vessels are the splenic (60%-80%), gastroduodenal (20%-25%), pancreaticoduodenal (10%-15%), hepatic (5%-10%) and left gastric (2%-5%) arteries [4]. Rupture of the splenic vein into the pancreatic duct has also been reported [6]. Following complicated pancreatitis, the next most common etiologies are primary vascular diseases (such as visceral aneurysms and arteriovenous malformations) [4, 7, 8] and tumors (such as pancreatic ductal adenocarcinoma, pancreatic neuroendocrine tumors (PNETs), and carcinomas in situ [9]). Even biopsy of such tumors during EUS has produced HP [10-12].

Angiography and surgery are the two main treatment options. Selective transcatheter embolization of the

bleeding artery or pseudoaneurysm is reserved for those patients who are hemodynamically stable and do not have another indication for resection. Many cases have been reported, with success rates ranging from approximately 80% to 100% [13-17]. Recurrent bleeding may occur in 10% to 37% of patients following embolization [17].

In this case, resection via distal (left) pancreatectomy was recommended, given evidence of a symptomatic and easily resectable lesion. Interestingly, although MRI, EUS, operative, and gross pathological findings were concordant with a bleeding PNET, viz., a lobular, discrete, firm, early-arterial- hyperenhancing mass at the tail of the pancreas, microscopic pathological evaluation revealed no neoplasm, only a nodule of pancreatic tissue containing a paucity of interlobular connective tissue and a relative loss of lobularity, imparting a pseudonodular appearance to the acinar and islet cells (Figures 4 and 5).

In conclusion, we present what we believe to be the first reported case of HS associated with noninflammatory pancreatic pseudonodule. Although it is possible that the HS may have been simply incidental to the nodule, and not

caused by the nodule, no other liver or pancreatic mass, vascular lesion, or evidence of pancreatitis was present to explain the HP.

Conflict of Interest Statement

The authors declare that they have no conflict of interest.

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