



Hemosuccus Pancreaticus: Diagnostic Pitfalls of a Rare Condition

Bradley Busebee, MD¹, Kurdi AT, MD², Stultz BR, MD¹, Sayegh LN, MD³, and Nayantara Coelho-Prabhu, MBBS²

¹Department of Internal Medicine, Mayo Clinic, Rochester, MN

²Department of Gastroenterology and Hepatology, Mayo Clinic, Rochester, MN

³Faculty of Medicine, American University of Beirut Medical Center, Beirut, Lebanon

ABSTRACT

The combination of cirrhosis and chronic pancreatitis is rare and poses increased risk of hemorrhage requiring close clinical monitoring. We present a patient with history of alcohol-associated cirrhosis and chronic pancreatitis who was admitted to the intensive care unit with clinical hemorrhage believed secondary to epistaxis. After initial delay, esophagogastroduodenoscopy ultimately found blood and clots evacuating through the ampulla consistent with hemosuccus pancreaticus confirmed with computed tomography angiography. The patient ultimately improved with coil and gel foam vascular embolization. This case highlights the dangers of early diagnostic closure and presents a rare finding of hemosuccus without pseudoaneurysm formation.

KEYWORDS: hemosuccus pancreaticus; gastrointestinal hemorrhage; cirrhosis; chronic pancreatitis; upper gastrointestinal bleed

INTRODUCTION

Cirrhosis and chronic pancreatitis are independently associated with an increased risk of hemorrhage. Comorbid cirrhosis and chronic pancreatitis is rare¹ and requires particularly careful evaluation for hemorrhagic complications. Among cirrhotic patients, the most common cause of hemorrhage is variceal bleeding, accounting for roughly 50%–78% of upper gastrointestinal bleeds. Peptic ulcer disease, Mallory-Weiss tear, portal hypertensive gastropathy, and erosive esophagitis account for the majority of the remaining case.^{2–4} In patients with acute pancreatitis, 80% of gastrointestinal bleeds are attributable to esophageal or gastroduodenal ulcers. However, nearly half of hemorrhages in patients with acute pancreatitis are extraluminal, with the majority of these involving hemorrhage into a pancreatic collection or pseudocyst,⁵ which may manifest in hemosuccus pancreaticus (HP). HP is an exceedingly rare cause of gastrointestinal hemorrhage and is characterized by bleeding from the duodenal papilla through the pancreatic duct.⁶ Here, we report an atypical case of HP in a patient with comorbid alcohol-associated cirrhosis and chronic pancreatitis without pseudoaneurysm formation.

CASE REPORT

A 31-year-old woman with decompensated alcoholic cirrhosis and chronic pancreatitis presented with hypovolemic shock in the setting of epistaxis and tertiary adrenal insufficiency. On admission, she required blood products, pressor support, and stress dose steroids. Her epistaxis resolved with these measures, and she was weaned off pressors with stable hemodynamics. Endoscopy was initially deferred because she had undergone endoscopy a month earlier, during a similar presentation of epistaxis and hematochezia, which showed mild portal hypertensive gastropathy without any luminal evidence or sources of bleeding or varices. A week after admission to the intensive care unit, she was transferred to the medical ward. Although she remained hemodynamically stable, the patient experienced sudden onset of abdominal pain and hematochezia. Repeat esophagogastroduodenoscopy demonstrated fresh blood and clots emanating from the ampulla (Figure 1). Computed tomography angiography revealed active arterial bleeding within the pancreatic head without pseudoaneurysm formation (Figure 1), consistent with HP, as well as acute on chronic pancreatitis with peripancreatic fluid collections in the head and tail. Hemostasis was achieved through interventional radiology-guided

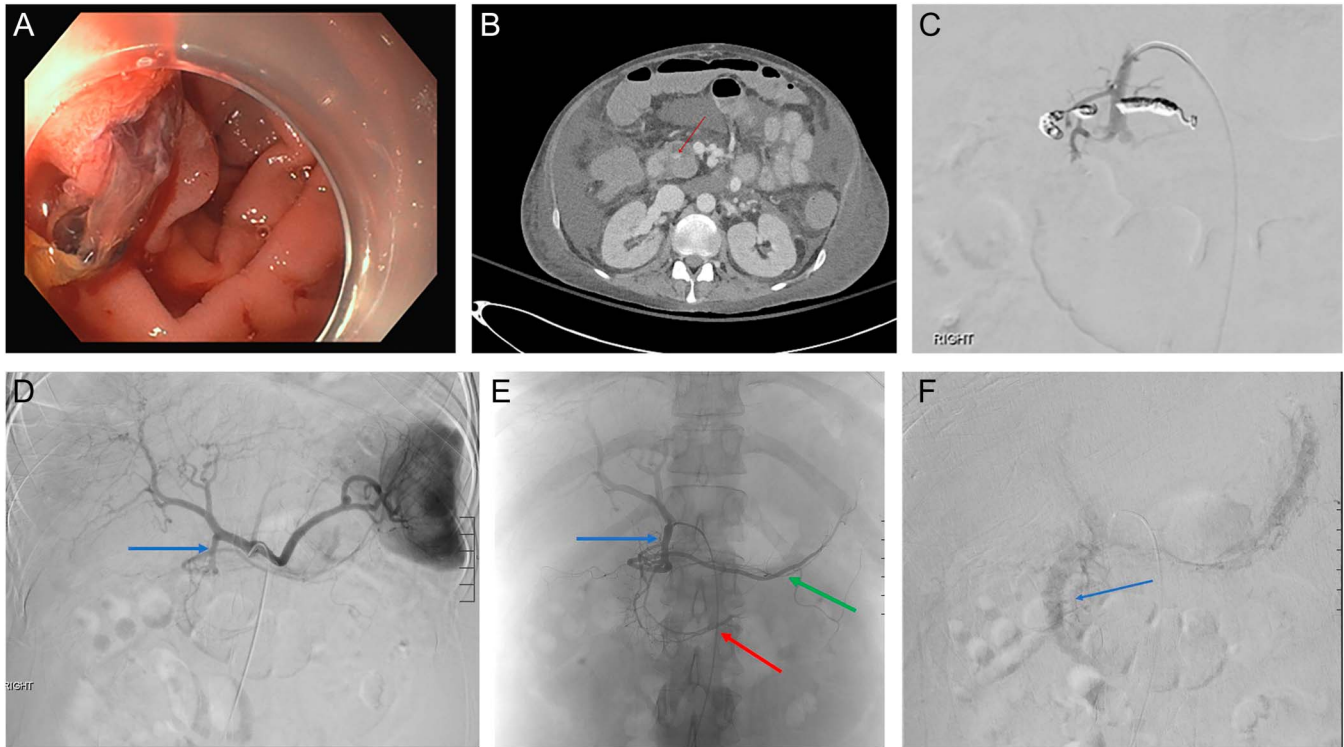


Figure 1. (A) Endoscopic visualization of the proximal duodenum with apparent fresh and clotted blood emanating for the ampulla. (B) Portal phase of a computed tomography angiography series showing a cyst within the head of the pancreas and accumulation of contrast within the cyst indicating active bleeding. (C) Coil embolization of the right gastroepiploic artery and gel foam embolization of the gastroduodenal artery and pancreaticoduodenal arcade. (D) Fluoroscopy showing injection of the celiac axis without evidence of associated aneurysm or pseudoaneurysm. The blue arrow indicates the gastroduodenal artery. (E) Fluoroscopy showing direct injection of the gastroduodenal artery without evidence of associated aneurysm or pseudoaneurysm. The blue arrow indicates the gastroduodenal artery, the red arrow the pancreaticoduodenal arteries, and the green arrow the transverse pancreatic artery. (F) Delayed fluoroscopy after injection of the gastroduodenal artery demonstrating contrast blush along the second part of the duodenum indicating active bleeding as indicated by the blue arrow.

coil embolization of the right gastroepiploic artery and gel foam embolization of the gastroduodenal artery and pancreaticoduodenal arcade (Figure 1).

DISCUSSION

Cirrhosis and chronic pancreatitis both increase the risk of numerous hemorrhagic processes. Although variceal bleeding is common and a major concern in patients with alcohol-related organ dysfunction, it is important to consider other hemorrhagic processes, particularly those related to any underlying pancreatic disease, including HP. Chronic pancreatitis is present in 76% of cases of HP, and acute pancreatitis is present in 13%.⁷ Other, rare etiologies of HP include neoplasm and trauma.⁶

There are a small number of case reports of HP in patients with cirrhosis,^{8–10} and one retrospective study showing that 1 of the 17 patients treated for HP at a single center had comorbid cirrhosis.¹¹ However, despite numerous overlapping risk factors for liver disease and chronic pancreatitis, there are no larger retrospective studies on the prevalence of comorbid liver disease and HP. It is not known whether the coagulopathy of cirrhosis, marked by decreased coagulant cofactor levels, endothelial dysfunction,

renal dysfunction, and portal hypertension,¹² increases the risk of HP itself.

Pancreatitis-induced inflammation and erosion likely promote vessel degradation and pseudoaneurysm formation. Although pseudoaneurysms are common in chronic pancreatitis,¹³ the majority of these never result in hemorrhage. Pseudoaneurysm rupture is the most common cause of HP, and as many as 98% of patients with HP have ≥ 1 pseudoaneurysm.¹⁴ Bleeding pseudoaneurysms most often involve the splenic artery and less commonly the gastroduodenal artery, pancreaticoduodenal arteries, and hepatic artery.^{7,15} A minority of patients with HP have no identifiable pseudoaneurysm,⁷ and absence of pseudoaneurysm on cross-sectional imaging, such as in this case, does not rule out HP.

HP may present as melena or hematochezia and less often hematemesis.¹⁴ Bleeding and abdominal pain are often intermittent, which may result from intermittent papillary duct obstruction from passing blood clots that may temporarily raise intraductal papillary pressures and prevent passage of blood into the small bowel lumen.¹⁶ Regardless of the pathophysiology, the intermittent nature of the bleeding likely explains the poor sensitivity of endoscopy, which is around

81%, and frequent delays in diagnosis.^{7,14} Optimizing the sensitivity of endoscopy also requires careful inspection of the papilla, which may be overlooked as a source for hemorrhage.

Although endoscopy may show bleeding from the papilla and helps to exclude other hemorrhagic etiologies, computed tomography angiography is the gold standard for diagnosing HP¹⁷ and should be considered in patients with pancreatic disease and suspicion of gastrointestinal hemorrhage. For these multiple reasons, a negative endoscopy does not preclude a diagnosis of hemosuccus. A low threshold is necessary for investigating possible HP because mortality is reported as high as 9.6%¹⁸ and may exceed 90% without adequate intervention.¹⁹ Endovascular and surgical management are effective in controlling hemorrhage and reducing mortality; in addition, recurrence of HP after effective treatment is rare.^{6,14}

This case of HP is notable for several atypical features, including the concurrence of alcohol-induced cirrhosis and pancreatitis, which are rarely comorbid but even more rare to manifest at such a young age. It also illustrates the importance of maintaining a low threshold for repeat endoscopy for patients with hemorrhagic shock, particularly in the setting of conditions that increase bleeding risk, regardless of negative index examination, which may not capture certain hemorrhagic processes that may manifest in intermittent and atypical arterial sources of bleeding. Furthermore, although HP typically develops from splenic and gastroduodenal arterial pseudoaneurysms, the arterial source of HP here was notably atypical and void of pseudoaneurysmal formation.

DISCLOSURES

Author contributions: B. Busebee wrote the manuscript and is the article guarantor. Kurdi AT and Stultz BR reviewed the manuscript. Sayegh LN performed background research and wrote and reviewed the manuscript. N. Coelho-Prabhu was the supervising physician, attending proceduralist, and reviewed the manuscript.

Financial disclosure: N. Coelho-Prabhu is a consultant to Boston Scientific and Alexion Pharma. The authors have no other conflicts of interest or study support items to disclose.

Informed consent was obtained for this case report.

Received September 29, 2022; Accepted February 21, 2023

REFERENCES

- Chand SK, Pendharkar SA, Bharmal SH, Bartlett AS, Pandol SJ, Petrov MS. Frequency and risk factors for liver disease following pancreatitis: A population-based cohort study. *Dig Liver Dis*. 2019;51(4):551–8.
- Christensen E, Fauerholdt L, Schlichting P, et al. Aspects of the natural history of gastrointestinal bleeding in cirrhosis and the effect of prednisone. *Gastroenterology*. 1981;81(5):944–52.
- Odelowo OO, Smoot DT, Kim K. Upper gastrointestinal bleeding in patients with liver cirrhosis. *J Natl Med Assoc*. 2002;94(8):712–5.
- Seo YS, Kim YH, Ahn SH, et al. Clinical features and treatment outcomes of upper gastrointestinal bleeding in patients with cirrhosis. *J Korean Med Sci*. 2008;23(4):635–43.
- Sharma PK, Madan K, Garg PK. Hemorrhage in acute pancreatitis. *Pancreas*. 2008;36(2):141–5.
- Yu P, Gong J. Hemosuccus pancreaticus: A mini-review. *Ann Med Surg*. 2018;28:45–8.
- Yashavanth HS, Jagtap N, Singh JR, et al. Hemosuccus pancreaticus: A systematic approach. *J Gastroenterol Hepatol*. 2021;36(8):2101–6.
- Amin H, Thomas M, Chand G, Mishoe M. 3002 Several endoscopies later, hemosuccus pancreaticus comes to light. *Am J Gastroenterol*. 2019;114(1):S1626.
- Bul V, Boulay B. Hemosuccus pancreaticus causing profound anemia in a Jehovah's witness with cirrhosis. *Am J Gastroenterol*. 2015;110:S79.
- Meena BL, Praharaj DL, Premkumar M, Mehtani R, Chaluvashtetty SB. Hemosuccus pancreaticus: Masquerader of variceal bleed in patient with alcoholic steatohepatitis. *Eur J Gastroenterol Hepatol*. 2022;34(2):235–6.
- Lermite E, Regenet N, Tuech JJ, et al. Diagnosis and treatment of hemosuccus pancreaticus: Development of endovascular management. *Pancreas*. 2007;34(2):229–32.
- Tripodi A, Mannucci PM. The coagulopathy of chronic liver disease. *N Engl J Med*. 2011;365(2):147–56.
- El Hamel A, Parc R, Adda G, Bouteloup PY, Huguet C, Malafosse M. Bleeding pseudocysts and pseudoaneurysms in chronic pancreatitis. *Br J Surg*. 2005;78(9):1059–63.
- Ru N, Zou WB, Qian YY, et al. A systematic review of the etiology, diagnosis, and treatment of hemosuccus pancreaticus. *Pancreas*. 2019;48(5):e47–9.
- Han B, Song ZF, Sun B. Hemosuccus pancreaticus: A rare cause of gastrointestinal bleeding. *Hepatobiliary Pancreat Dis Int*. 2012;11(5):479–88.
- Cui H-Y, Jiang CH, Dong J, Wen Y, Chen YW. Hemosuccus pancreaticus caused by gastroduodenal artery pseudoaneurysm associated with chronic pancreatitis: A case report and review of literature. *World J Clin Cases*. 2021;9(1):236–44.
- Tarar ZI, Khan HA, Inayat F, et al. Hemosuccus pancreaticus: A comprehensive review of presentation patterns, diagnostic approaches, therapeutic strategies, and clinical outcomes. *J Invest Med High Impact Case Rep*. 2022;10:232470962110703.
- Maheshwaran MU, Sathyanesan J, Ramasamu S, et al. Hemosuccus pancreaticus: 18-year experience from a tertiary care GI bleed centre in India. *IHPBA*. 2016;18:e784.
- Carr JA, Cho JS, Shepard AD, Nypaver TJ, Reddy DJ. Visceral pseudoaneurysms due to pancreatic pseudocysts: Rare but lethal complications of pancreatitis. *J Vasc Surg*. 2000;32(4):722–30.

Copyright: © 2023 The Author(s). Published by Wolters Kluwer Health, Inc. on behalf of The American College of Gastroenterology. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.