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Case Report

Diagnostic and Therapeutic Challenges in Hemosuccus Pancreaticus: A Case Report*,**

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ARTICLE INFO

Article history: Received 5 August 2024 Revised 16 September 2024 Accepted 17 September 2024

Keywords:
Hemosuccus pancreaticus
Gastrointestinal bleeding
Pseudoaneurysm
CT angiography
Embolization
Arteriography

ABSTRACT

Hemosuccus pancreaticus, also known as pancreatic pseudoaneurysm, is a rare type of upper gastrointestinal bleeding that originates from the major duodenal papilla or ampulla of Vater and extends into the duodenum via the pancreatic duct. By 2021, only 150 cases had been reported worldwide. Without treatment, the mortality rate can be as high as 90%, but it drops to 25%-37% with intervention. We report a case of a 64-year-old woman who experienced intermittent upper gastrointestinal bleeding over a year. Imaging revealed a hematoma and pseudoaneurysm in the pancreas, which was successfully treated with angiographic embolization. This case highlights the value of a multidisciplinary approach, using advanced imaging and minimally invasive techniques like embolization for effective patient outcomes.

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Introduction

Hemosuccus pancreaticus (HP) also known as pancreatic pseudoaneurysm, is defined as bleeding originating from the major duodenal papilla, ampulla of Vater, and extending into the second portion of the duodenum through the pancreatic duct [1,2]. It is a rare clinical event of upper gastrointestinal bleeding; up until 2021, only 150 cases of HP had been reported worldwide [3]. It is a clinical complication that occurs in pa-

tients with a history of acute (3.5%) or chronic (10%) pancreatitis, pancreatic tumor, pancreas divisum, pancreatic pseudocyst, and iatrogenic lesion of the pancreas [4]. It occurs in approximately one out of every 1500 cases. The male-to-female ratio is 7:1, and it typically occurs between the fifth and sixth decades of life. In untreated cases, the mortality rate can reach 90%, while in treated cases, it varies between 25% and 37% [5].

Among the clinical characteristics of this pathology, it may present with symptoms of intermittent upper or

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https://doi.org/10.1016/j.radcr.2024.09.099

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 $^{^{\}scriptsize{\pm}}$ Competing Interests: The authors have declared that no competing interests exists.

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lower gastrointestinal bleeding such as melena, hematochezia, hematemesis, abdominal pain, and hypovolemic shock. Additionally, chronic anemia may be evident [6]. The diagnostic and therapeutic approach is 1 of the main challenges of this pathology due to its anatomic origin and the frequent negative results in upper endoscopic studies. This is why computed tomography (CT) angiography is considered the gold standard for identifying the bleeding site and selecting the ideal therapeutic intervention [6].

We present the case of a 64-year-old female patient with a one-year history of recurrent gastrointestinal bleeding, with evidence on CT angiography of a hematoma with a pseudoaneurysm in the body and tail of the pancreas, associated with thrombosis of the superior mesenteric vein and the splenic vein.

Case Report

A 64-year-old female patient presented with a 1-year history of upper gastrointestinal tract hemorrhage. Her medical history includes arterial hypertension, type 2 diabetes mellitus, and cholecystitis with pancreatitis, whom required cholecystectomy and percutaneous drainage of a pancreatic pseudocyst 2 years ago. On physical examination, she was in fair general condition, albeit with hypotension and generalized mucocutaneous pallor. No abdominal alterations or evident active bleeding were observed.

Treatment was initiated with intravenous fluids and blood transfusions for anemia, which successfully stabilized her hemodynamically. An abdominal CT angiography revealed a hematoma with an internal pseudoaneurysm, occupying the body and tail of the pancreas. The hematoma had an approximate volume of 150 cc. Within this hematoma, a pseudoaneurysm measuring 31×17 mm (transverse x AP) was observed, connected to a pancreatic branch originating from the splenic artery. Despite the portal vein being patent, thrombosis of the superior mesenteric vein and splenic vein was identified, resulting in multiple peri-splenic and peri-gastric collaterals (Fig. 1). No signs of active bleeding were observed on image (Fig. 2).

Procedure

Embolization of the pancreatic pseudoaneurysm was performed using the trapping technique. An aortogram was conducted to identify the visceral branches. The celiac trunk was identified, giving rise to the left gastric artery, common hepatic artery, and splenic artery. Catheterization was challenging due to the anatomical angle of the celiac trunk and splenic artery. Arteriography of the splenic artery revealed an aneurysm measuring 36×32 mm, emerging from an early and inferior branch of the splenic artery (Fig. 3).

It had a 1.5 mm neck and originated from the inferior margin very close to the main splenic artery. The branch from which the pseudoaneurysm originated was catheterized with an SL 10 microcatheter and Synchro Microguide and em-

bolized distally with coils using the trapping technique. Embolization was completed with a 50% Histoacryl-Lipiodol mixture. At the end, the patency of the other vascular structures of the spleen was preserved (Figs. 4 and 5).

The patient had a satisfactory recovery, with no new episodes of gastrointestinal bleeding, and follow-up bloodwork showed an increase in hemoglobin. Consequently, the patient was discharged and continues to be monitored by gastroenterology and internal medicine.

Discussion

Hemossuccus pancreaticus (HP) is a very rare presentation of upper gastrointestinal bleeding, also known as pseudohemobilia or Wirsungorrhagia [7]. It is characterized by intermittent bleeding originating from the major duodenal papilla and extending into the second portion of the duodenum through the pancreatic duct [1]. On average, symptoms persist for more than 30 days before HP is diagnosed [7].

This pathology can occur secondary to various etiologies, including pancreatitis [1]. Pancreatitis, which can be acute, chronic, or hereditary, is responsible for 80% of HP cases, causing inflammatory changes at the level of the pancreatic ducts. Pancreatic enzymes, such as elastase, can rupture the vascular wall, leading to cyst formation, which occurs in 10% to 17% of cases of chronic pancreatitis and cholelithiasis [8]. Communication between the cyst and the artery results in a pseudoaneurysm [3], as likely occurred in this patient's case.

The most common site for HP formation is the splenic artery, while the least common site is the gastroduodenal artery. However, it can also occur in the superior mesenteric artery, celiac trunk, cystic artery, right hepatic artery, and inferior mesenteric artery [3]. To date, no specific relationship has been described between the different etiologies of HP and the type of artery affected.

In addition, HP can result from procedural complications, such as the drainage of a pancreatic pseudocyst [1]. In the case of our patient, she had history of pancreatitis and cholelithiasis 2-years ago, and underwent percutaneous drainage of a pancreatic pseudocyst, all of which are possible etiologies of HP. This suggests that HP is a multifactorial clinical entity, making it crucial to diagnose and treat all factors that increase the risk of its formation. The diagnosis of HP is complicated by its infrequency, its anatomical location, the intermittent nature of its symptoms, and the multiple causes that can provoke it [9], making it an underdiagnosed pathology. Laboratory tests are usually normal and inconclusive, so imaging is the diagnostic method of choice [4].

Upper endoscopy is the preferred test for diagnosing upper gastrointestinal bleeding, it can detect bleeding in only approximately 30% of cases of HP. Therefore, a negative endoscopy does not rule out the possibility of HP, as the forward-viewing endoscope may have an intermittent and suboptimal view of the ampulla of Vater. Moreover, bleeding in this pathology is typically intermittent [2,6]. On the other hand, ERCP (endoscopic retrograde cholangiopancreatography) uses a side-viewing endoscope that allows for visualization of the pancreatic duct and the source of bleeding. A retrospec-

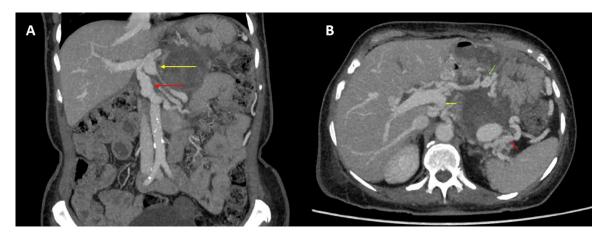


Fig. 1 – Image (A) Yellow arrow: Mesenteric vein thrombosis. Red arrow: Collateral venous circulation. Image (B) Perigastric collateral circulation (green arrow) and perisplenic collateral circulation (red arrow). Yellow arrow: Splenic vein thrombosis.



Fig. 2 – The pseudoaneurysm (yellow arrow) measures 31×17 mm (transverse x AP). This pseudoaneurysm originates from a branch of the splenic artery.

tive study reported a diagnostic success rate of 29.6% with conventional endoscopy compared to 70.1% with side-view endoscopy [10].

Computed tomography (CT) can show positive results in 90% of cases and can be performed using multiphase CT angiography or CT enterography. This type of imaging may reveal signs suggestive of HP, such as blood clots within the pancreatic duct, known as 'sentinel clot signs', continuous opacification within the aneurysms, presence of pseudocysts, or persistent contrast enhancement beyond the arterial phase [6].

Angiography is the diagnostic method of choice that allows identification of the affected artery, delineation of the arterial anatomy, and therapeutic intervention [11]. For managing this condition, arterial embolization is the first-line treatment, with success rates ranging from 79% to 100% [6]. However, surgical management is also an option, though it is associated with higher mortality rates and a risk of rebleeding [4].

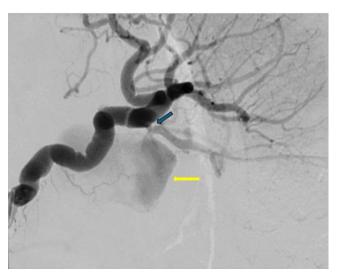


Fig. 3 – The pseudoaneurysm (yellow arrow) is observed emerging from an inferior branch of the splenic artery (blue arrow).

The choice of treatment depends on the patient's clinical condition. In hemodynamically stable individuals, such as in the case of this patient, selective transcatheter embolization of the bleeding artery or pseudoaneurysm is indicated, provided there is no other etiology requiring surgical management [3]. Embolization techniques include stenting, balloon tamponade, and the use of prosthetic materials [2]. Materials that may be used include n-butyl 2-cyanoacrylate, gelatin sponge, thrombin, and coils, with coils being the most employed [6,12]. During this procedure, coils are deployed to fill the pseudoaneurysm, inducing complete thrombosis, followed by closure of the opening. The affected artery is completely embolized at its proximal and distal ends to isolate the pseudoaneurysm from the blood supply, thereby achieving

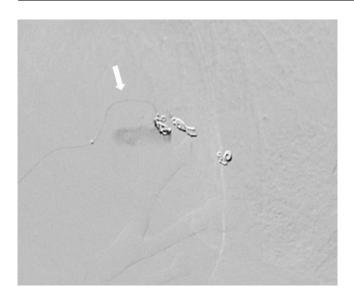


Fig. 4 – Selective catheterization with microcatheter and embolization using the trapping technique (white arrow).

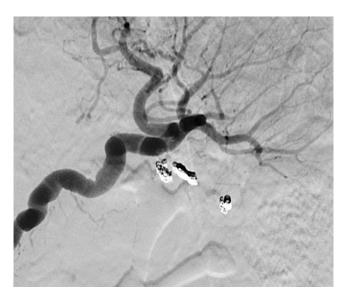


Fig. 5 – Final angiographic control, exclusion of the pseudoaneurysm.

hemostasis. Following this procedure, the risk of rebleeding is 37% [6].

Our patient underwent angiographic embolization. Despite challenges in catheterization due to the anatomical angle of the celiac trunk and splenic artery, and the proximity of the inferior margin to the main splenic artery, the patient had a favorable outcome and did not experience rebleeding.

On the contrary, surgical treatment is indicated in patients with uncontrolled bleeding, hemodynamic instability, persistent shock, or when intervention by interventional radiology fails due to recurrent or continuous bleeding. Despite the low rebleeding rate (between 0% and 5%), surgical management carries a mortality rate of 20%-25% [13].

Conclusion

Haemosuccus pancreaticus is a rare and complex diagnostic entity that manifests as intermittent and potentially severe gastrointestinal bleeding. The case of a 64-year-old female patient described here illustrates the diagnostic and therapeutic challenges associated with this condition.

This case underscores the importance of a multidisciplinary approach to managing HP, integrating advanced imaging techniques such as multiphase CT and angiography with minimally invasive therapeutic interventions like arterial embolization. The patient's favorable outcome, characterized by the absence of new bleeding episodes and improvement in hematological parameters, highlights the effectiveness of this approach.

It is crucial to consider all potential etiologies and employ accurate diagnostic methods to identify and appropriately address risk factors. Arterial embolization, with its high success rate and lower risk of complications compared to surgery, is established as the primary treatment for hemodynamically stable patients. This case provides valuable evidence of effective HP management.

Patient consent

The reported case was reviewed and approved, and individual patient consent was obtained following institutional guidelines.

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