

***Hemosuccus Pancreaticus* as a Rare Cause of Gastrointestinal Bleeding: a Report of Two Cases**

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Abstract- Pancreatic diseases are known to be associated with complications such as pseudocyst and abscess. A pseudoaneurysm associated with pancreatitis may develop as well. The pseudoaneurysm may rupture into various parts of the gastrointestinal tract; the peritoneal cavity, or the retroperitoneum. We report two cases of *Hemosuccus pancreaticus* admitted to our center in the past five years. One case was associated with acute pancreatitis, and another case was associated with chronic pancreatitis. A pseudocyst was found in two cases. Both were successfully managed by emergency surgery. So, *Hemosuccus pancreaticus* is a rare cause of gastrointestinal bleeding with difficult diagnosis, and surgery is the effective treatment for the patient with severe bleeding. However in cases with no life-threatening bleeding angiography and embolization can be performed.

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Introduction

Pancreatic diseases are known to be associated with complications such as pseudocyst and abscess formation (1,2). Pancreatic hemorrhage as a cause of massive gastrointestinal hemorrhage is a rare event (3) but its importance due to high mortality rate (4) makes us well aware of this condition. Causes of relation between the vascular and biliary or pancreatic system include trauma, pancreatitis, liver biopsy, gallstone, aneurysms in hepatic or gastroduodenal arteries, hepatic abscess, chronic alcoholism and neoplasm (2,5).

Patients are classically presented with upper gastrointestinal (GI) bleeding accompanied by biliary colic and jaundice. The diagnosis is made by endoscopic visualization of blood coming from the ampulla of Vater and/or by angiography (5,6).

The ampullary bleeding also may originate from the pancreatic duct; this condition is termed as *Hemosuccus pancreaticus* (7). Large cysts almost invariably cause compression of the adjacent stomach or duodenum. Smaller cysts in the head of the pancreas make the proximal duodenum fix and narrow, whereas those located in the tail may produce deformity in the posterior gastric wall near to the esophagogastric

junction (2). Most patients have chronic pancreatitis with either erosion of a pseudocyst into a peripancreatic artery or formation of an arterial aneurysm that may subsequently develop and make a way to the pancreatic duct (8). A pseudoaneurysm associated with pancreatitis may be developed either by self-digestion of a vessel wall by proteolytic enzymes released from the pancreas or by direct pressure erosion from an adjacent pseudocyst. The involved arteries in pseudoaneurysm formation are the splenic artery followed by the gastroduodenal artery and the pancreatic duodenal arteries.

Aneurysms of the hepatic, celiac, superior mesenteric and gastric arteries have also been reported. The pseudoaneurysm can rupture into various parts of the gastrointestinal tract, the peritoneal cavity, or the retroperitoneum (9). Such hemorrhages usually result from inflammatory or enzymatic erosions in adjacent viscera vessels with the formation of an expanding false aneurysm and subsequent rupture into the stomach or duodenum (10). This entity is also known as *Hemosuccus pancreaticus*, hemoductal pancreatitis, splenic artery (aneurysm) pancreatic duct fistula and Wirsungorrhage or hemowirsungia (11).

This report elucidates two cases of upper

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gastrointestinal bleeding secondary to pancreatic pseudocyst dislocation.

Case Report

Case 1

A 33-year-old female presented with severe epigastric pain. Her medical history indicated cholecystectomy one week before admission. A computed tomography (CT) scan revealed pancreatitis and a pseudocyst of 95×70 mm in size (Figure 1).

The patient underwent laparotomy and cystogastrostomy. Twelve hours later she experienced hematemesis. Because the patient continued active bleeding and required multiple blood transfusions, an emergency surgery was performed immediately.

Upon entering the abdomen and after opening the anterior gastric wall, fresh blood was noted. Active bleeding was observed at the site the pseudocyst. Exploration of the hemorrhagic site revealed bleeding from the gastroduodenal artery. Bleeding was controlled by oversewing. Patient was discharged 10 days after surgery.



Figure 1. Contrast abdominal computed tomography scan showing pancreatic pseudocyst

Laboratory tests showed amylase: 305 U/L, lipase: 520 U/L, total bilirubin: 0.6 mg/dL aspartate aminotransferase: 15 IU/L, alanine aminotransferase: 141 IU/L, and alkaline phosphatase: 98 IU/L

Case 2

The patient was a 54-year-old man with a history of massive GI bleeding who admitted to the emergency department.

The patient vital signs were: BP: 90/60 mmHg, PR: 145/min, T: 37.5 °C and RR: 24/min.

Resuscitation was done with Ringer lactate serum (2 liters), FFP (5 units), packed red blood cell (4 units).

Ranitidine (50 mg) and gastric lavage with cold serum N/S also were administrated.

Examinations revealed that the patient was pale with signs of confusion through 4-5 hrs after admission. Bleeding was continued, and there was a life threatening situation.

Initial laboratory tests revealed RBC count: $4.09 \times 10^6/\text{ml}$, Hgb: 10 g/dl, Plt: $253 \times 10^3/\text{ml}$, Hct: 30%, K^+ : 5.3 meq/ml, Na^+ : 130 meq/ml, serum creatinine: 0.8 mg/dl, Urea: 28 mg/dl, and blood sugar: 148 mg/dl. The repeated CBC after 3 hrs of admission shows: Hct: 23%, Hgb: 7.5 g/dl, RBC count: $2.94 \times 10^6/\text{ml}$, Plt: $112 \times 10^3/\text{ml}$.

The results of arterial blood gas (ABG) analysis demonstrated: pH: 7.23, HCO_3^- : 15 meq/L, PCO_2 : 37 mmHg, PO_2 : 54 mmHg and O_2Sat : 55%.

Patient history revealed hospitalization due to the pancreas pseudocyst two weeks earlier. The patient underwent an open cystogastrostomy via the posterior approach and was discharged two days afterward.

Patient was unstable during first 5 hrs of admission. He was transferred to the operating room due to or because of severe gastrointestinal bleeding. Reanimation was continued with packed cell (4 units), FFP (5 units), Ringer's lactate as fluid resuscitation (2 liters).

The abdomen was opened through the same previous midline abdominal wall incision. Abdomen cavity was cleaned out; no sign of hemorrhage or fibrin was observed. Further exploration showed a new pseudocyst in greater curvature with the dimension of 15×20 cm. After opening the anterior stomach wall and the blood clots were removed manually. Fresh blood was also present and was evacuated or aspirated using a suction device.

Active bleeding was observed in the posterior gastric wall and at the site of previous pseudocyst. Gastroduodenal artery bleeding was controlled by oversewing, and gastrojejunostomy treated the cyst.

During the operation, patient received 10 units of packed cells and was discharged from the hospital 10 days after admission. The patient was done well, with no further evidence of bleeding during nine-month follow-up.

Discussion

Pancreatic pseudocysts are fluid collections arising from tissue within and around the pancreas with the lacking an epithelial lining (13). They can occur following acute pancreatitis, chronic pancreatitis or

secondary to pancreatic trauma (4,14). Pancreatic pseudocysts which are arising after acute pancreatitis can be managed conservatively usually by drainage (15).

It is suggested in Baron and Morgan study that pseudocysts which have not regressed or have increased in size after 6 weeks is better to be drained surgically (16).

Although it is a rare event, but spontaneous gastrointestinal bleeding in a patient with pancreatitis and a demonstrable or suspected pseudocyst demands both a high degree of clinical awareness about the possibility of a false aneurysm, and a defined diagnostic and therapeutic stratagem (10).

Endoscopy is mandatory to exclude one of the more common causes of upper gastrointestinal bleeding, and should visualize the second part of the duodenum with a side endoscopic imaging for optimal papillary views (3).

Blood issuing from the papilla is a diagnostic criterion for pancreatic duct hemorrhage, whereas the absence of an alternative cause is strongly suggestive (5,10).

Arteriography with selective celiac axis and superior mesenteric imaging is the definitive investigation (6,10,12).

In present cases, endoscopic evaluation and arteriography were not performed because of continuous massive hemorrhage which required emergency surgery.

Surgical therapy consists of either pancreatic resection or suture ligation of the involved vessels (10,11).

Predictably, resection mortality rate depends on the site of bleeding (11). Distal pancreatectomy and pancreatic head resection are associated with 16 and 43 % of mortality, respectively (10).

Although endoscopy and embolization angiography are two diagnostic and treatment options in patients with upper GI bleeding, which we did not take advantage of due to severe hemorrhage. Under this condition, angiography may demonstrate hemorrhage from the site other than the exact one (1).

Massive bleeding through the duct of Wirsung because of breakage of a splanchnic artery pseudoaneurysm into a pancreatic pseudocyst is one of the of hemoductal pancreatitis etiologies (7). Surgical management of hemorrhagic pseudocysts includes excision of the pseudoaneurysm and pseudocyst. If resection is impossible, ligation of the artery proximal and distal to the pseudoaneurysm and drainage of the pseudocyst into the gastrointestinal tract is an acceptable alternative, however it is associated with a higher re-bleeding rate (4,17).

While it is an uncommon cause of upper

gastrointestinal hemorrhage, spontaneous hemorrhage complicating pseudocyst formation was described in 6% of 112 cases by Sankaran and Walt and in 8% of 131 cases by Frey (10).

The unusual propensity for pancreatic pseudocysts to be associated with false aneurysms suggests that pancreatic enzymes play a pathogenic role (8).

Pseudocysts have been shown to contain activated lytic enzymes. Elastases are defined, in particular. These enzymes exert an elastolytic action on the elastic component of vessel walls and result in bleeding (10,18).

A pancreatic pseudocyst possesses a significant relation with the pancreatic duct, bleeding into the gastrointestinal tract may also occur entirely through this route (10).

The bleeding site most often has been the spleen artery, which besides the gastroduodenal and pancreatic duodenal arteries (5,9), constitute 78% of vessels; identified as being responsible for severe bleeding (6). Conservative management alone had 90% mortality, and surgical treatment showed 29% of mortality and for patients with arterial embolization, high mortality rate was reported (12).

In conclusion, *Hemosuccus pancreaticus* is a rare cause of gastrointestinal bleeding with a difficult diagnosis is. In the case of the pancreas pseudocyst, this rare complication should be considered in the differential diagnosis. Since bleeding is a life-threatening condition in these patients, it requires prompt diagnosis and timely planning for emergency operation. However, in cases with no life-threatening bleeding angiography and embolization can be performed. Oversewing the artery could be a safe and effective treatment for the patient with severe bleeding.

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References

1. Spivak H, Galloway JR, Amerson JR, et al. Management of Pancreatic Pseudocysts. J Am Coll Surg 1998;186:507-11.
2. Igari K, Ochiai T, Aihara A, et al. Hemosuccus pancreaticus caused by a primary splenic artery aneurysm as a rare cause of gastrointestinal bleeding: report of a case. Int Surg 2010;95(4):325-8.
3. Tsutsumi Y, Kunieda Y, Yamauchi J, et al. A case of

- hemosuccus pancreaticus caused by rupture of an aneurysm of the splenic artery. *Nihon Shokakibyō Gakkai Zasshi* 2013;110(7):1296-303.
4. Garcea G, Krebs M, Lloyd T, et al. Haemorrhage from Pancreatic Pseudocysts Presenting as Upper Gastrointestinal Haemorrhage. *Asian J Surg* 2004;27(2):137-40.
 5. Khalid A, Brugge W. ACG practice guidelines for the diagnosis and management of neoplastic pancreatic cysts. *Am J Gastroenterol* 2007;102(10):2339-49.
 6. Pinarbaşı B, Poturoğlu S, Yanar H, et al. A rare cause of hemosuccus pancreaticus: primary splenic artery aneurysm ruptured into pancreatic serous cystadenoma. *Turk J Gastroenterol* 2008;19(1):57-63.
 7. Grisendi A, Lonardo A, Della Casa G, et al. Hemoductal pancreatitis secondary to gastroduodenal artery-ruptured pseudoaneurysm: a rare cause of hematemesis. *Am J Gastroenterol* 1991;86(11):1654-7.
 8. Maeda A, Kunou T, Saeki S, et al. Pseudoaneurysm of the cystic artery with hemobilia treated by arterial embolization and elective cholecystectomy. *J Hepatobiliary Pancreat Surg* 2002;9(6):755-8.
 9. Chiu HH, Chen CM, Wang KCh, et al. Pancreatic pseudocyst bleeding associated with massive intraperitoneal hemorrhage. *Am J Surg* 2006;192(1):87-8.
 10. Ray S, Das K, Ray S, et al. Hemosuccus pancreaticus associated with severe acute pancreatitis and pseudoaneurysms: a report of two cases. *JOP* 2011;12(5):469-72.
 11. Han B, Song ZF, Sun B. Hemosuccus pancreaticus: a rare cause of gastrointestinal bleeding. *Hepatobiliary Pancreat Dis Int* 2012;11(5):479-88.
 12. Okuno A, Miyazaki M, Ito H, et al. Nonsurgical management of ruptured pseudoaneurysm in patients with hepatobiliary pancreatic diseases, 2001. *Am J Gastroenterol* 2001;96(4):1067-71.
 13. Grace PA, Williamson RC. Modern management of pancreatic pseudocysts. *Br J Surg* 1993;80(5):573-81.
 14. Warshaw AL. Pancreatic cysts and pseudocysts: new rules for a new game. *Br J Surg* 1989;76(6):533-4.
 15. Mullins RJ, Malagoni MA, Bergamini TM, et al. Controversies in the management of pancreatic pseudocysts. *Am J Surg* 1998;155(1):165-72.
 16. Baron TH, Morgan DE. The diagnosis and management of fluid collections associated with pancreatitis. *Am J Med* 1997;102(6):555-63.
 17. Rantala A, Ovaska J. Major hemorrhage associated with a pseudocyst in chronic pancreatitis: a gastro-surgical challenge. *Ann Chir Gynaecol* 1996;85(3):217-21.
 18. Somani PO, Jain SS, Shah DK, et al. Uncomplicated spontaneous ruptures of pancreatic pseudocyst into stomach: A case report. *World J Gastrointest Endosc* 2013;5(9):461-4.