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Hemosuccus Pancreaticus following a Puestow Procedure in a Patient with Chronic Pancreatitis

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Key Words

Hemosuccus pancreaticus · Puestow procedure · Chronic pancreatitis · Intraoperative endoscopy

Abstract

Hemosuccus pancreaticus is an unusual cause of gastrointestinal bleeding that occurs as a complication of chronic or acute pancreatitis. We report a case of extremely acute-onset hemosuccus pancreaticus occurring in a patient with chronic pancreatitis over a long-term follow-up after a Puestow procedure (side-to-side pancreaticojejunostomy). The patient was admitted to our hospital due to severe anemia and tarry stools indicative of gastrointestinal bleeding. Emergent endoscopy, including gastrointestinal fiberoscopy and colon fiberoscopy, showed no abnormal findings. Abdominal contrast-enhanced computed tomography and hemorrhagic scintigraphy did not detect a hemorrhagic lesion. Although interventional radiology was considered for diagnosis and treatment, conservative therapy seemed sufficient to affect hemostasis. Two weeks later, however, acute intestinal bleeding with hemodynamic shock occurred, and exploration was performed without delay. Intraoperative endoscopy through an incision of the reconstructed jejunal loop in the close proximal end revealed a site of active bleeding from the side-to-side anastomotic pancreatic duct. Following a longitudinal incision of the jejunal loop, a bleeding point was sutured and ligated on direct inspection. The patient showed a good postoperative course.

Introduction

Hemosuccus pancreaticus is known to be a rare cause of intermittent upper gastrointestinal bleeding from the duodenal papilla through the pancreatic duct. In

1931, Lower and Farrel first reported that blood was expelled into the duodenum via the main pancreatic duct [1]. The term 'hemosuccus pancreaticus' was coined by Sandblom in 1970 [2]. Reported causes of hemosuccus pancreaticus include not only benign pancreatic diseases, such as acute and/or chronic pancreatitis, pancreatic cysts and pseudoaneurysms, but also pancreatic neoplasms [3].

The diagnosis of hemosuccus pancreaticus is often difficult because the condition involves intermittent gastrointestinal bleeding. In our case, the cause of acute gastrointestinal bleeding was difficult to diagnose, due mainly to a combined history of longitudinal side-to-side pancreaticojejunostomy (Puestow procedure) [4]. Herein, we report an extremely rare case of hemosuccus pancreaticus in a long-term follow-up for chronic pancreatitis after a Puestow procedure. Furthermore, an active bleeding site could be directly inspected by intraoperative endoscopy. To our knowledge, this report is the first description of such a case in the literature, and we discuss the pathogenesis of this disease with a review of the relevant literature.

Case Report

A 47-year-old Japanese male was admitted to our hospital in November 2000 with severe anemia and tarry stools. The patient had a past history of peritoneal drainage for acute necrotizing pancreatitis at 23 years of age and a lateral pancreaticojejunostomy (Puestow procedure) for chronic pancreatitis at age 31. He experienced several episodes of tarry stools following the Puestow operation. Physical examination on admission revealed pallor conjunctiva and tarry stools. Laboratory data revealed the following lab values: RBC $181 \times 10^9/\mu\text{l}$; hemoglobin 4.2 g/dl; hematocrit 14.1%; WBC $4.8 \times 10^3/\mu\text{l}$; platelet count $220 \times 10^3/\mu\text{l}$; sodium 142 mEq/dl; chloride 105 mEq/dl; total protein 6.0 g/dl; albumin 3.77 g/dl; total bilirubin 1.04 mg/dl; alkaline phosphatase 127 U/l; LDH 127 U/l; GOT 29 U/l; GPT 44 U/l; serum amylase 39 U/l; elastase-1 110 ng/dl; lipase 78 U/l.

Emergent gastrointestinal fiberoscopy was performed, and no abnormal lesions were found in the stomach or duodenum. Hemorrhagic scintigraphy and abdominal computed tomography were also carried out, but no hemorrhagic lesions were found. After blood transfusion, total colonoscopy was performed via insertion up to the terminal ileum, which did not reveal abnormal lesions either. Although interventional radiology was considered as a treatment option, hemostasis seemed possible through conservative therapy alone. We presumed that the bleeding point was in the upper jejunum, including the reconstructed jejunum.

Conservative treatment for several days, including several packed red blood cell transfusions, improved the patient's anemia as well as his general condition; he began to eat shortly thereafter. Two weeks later, more tarry stools occurred, and the patient's hemoglobin level suddenly decreased to 5.0 g/dl. At this point, he went into hemodynamic shock and subsequently underwent laparotomy without delay. After inspection of the whole abdomen, the intestine was found to contain coagulated blood in the upper jejunum. Intraoperative endoscopy through an incision in the reconstructed jejunal loop at the close proximal end revealed massive bleeding (fig. 1a). After suction of the bleeding and coagulation clots, an active bleeding site from the side-to-side anastomotic pancreatic duct became clear (fig. 1b, c). Longitudinal jejunostomy was performed, and the bleeding point was sutured and ligated. Intraoperative findings from the opened pancreaticojejunostomy are shown in fig. 2a and b. In preparation for re-hemorrhage from the pancreaticojejunostomy site, a percutaneous jejunostomy was made in the left upper abdomen.

The patient's postoperative course was uneventful. A month later, an endoscopy was carried out via the prepared jejunostomy prior to his discharge from the hospital. The ligated site was observed and complete hemostasis was confirmed (fig. 2c). Six months later, the percutaneous jejunostomy was closed. It has now been ten years since the operation, and the patient is alive and without recurrence.

Discussion

Intermittent gastrointestinal bleeding of unknown cause is often a diagnostic challenge during routine clinical work. Hemosuccus pancreaticus is the term coined to describe the syndrome of gastrointestinal bleeding into the pancreatic duct manifested by blood loss through the ampulla of Vater [1, 2]. Our recent experience with a patient who had acute intermittent gastrointestinal blood loss was difficult to diagnose as hemosuccus pancreaticus because the patient had previously received a Puestow procedure, resulting in masking of a bleeding site of the reconstructed jejunal afferent loop. This is an extremely rare case of hemosuccus pancreaticus following a Puestow procedure.

Hemosuccus pancreaticus has been shown to be the cause of underlying chronic pancreatitis in more than 80% of pancreatic diseases [3]. Other pancreatic causes of hemosuccus pancreaticus are rare and include neuroendocrine tumors [5], ectopic pancreas [6], pancreas divisum [7], and intraductal papillary-mucinous carcinomas [8]. In chronic pancreatitis, this disease is usually caused by the rupture of a pseudoaneurysm in a peripancreatic artery to the pancreatic duct or by hemorrhage of a peripancreatic artery into the pseudocyst communicating with the pancreatic duct. In our case, abdominal computed tomography examination did not reveal a pancreatic cyst or a pseudoaneurysm in a peripancreatic artery. Operative findings showed that a hemorrhagic point was identified as an active crescendo-decrescendo arterial bleeding site via an opened pancreatic duct from the incised, lifted jejunum. Hemostasis was accomplished via direct suture ligation. Determination of the exact pathogenesis was complicated by postoperative vascular modifications, such as angiogenesis and vasculogenesis, following the Puestow procedure. It has been reported that the cause of hemosuccus pancreaticus can be a microrupture of the splenic artery with arteriosclerosis, as determined by histopathological examination of the resected specimen [9]. Peripancreatic microarterial changes such as pseudoaneurysms related to chronic pancreatitis were postulated to be responsible for the lesion.

There have been many reports concerning the effectiveness of interventional radiological therapy [10–13]. In the present case, angiography could not be performed because of the patient's emergent hemodynamic pre-shock condition. Surgery is the only option for the treatment of hemosuccus pancreaticus when angiography shows no abnormal findings and interventional radiological therapy is not successful. Surgical resection is an alternative treatment for hemosuccus pancreaticus, although in our case resection of the responsible lesion was not chosen because the patient's hemodynamic condition was unstable and a brief, simple operation was needed.

Intraoperative ultrasonography and pancreatoscopy have been reported to be useful tools to confirm the source of gastrointestinal bleeding and determine the cutting line of the pancreas [14]. In our case, intraoperative endoscopy was useful to confirm the origin of the bleeding. When emergent surgery is unavoidable in a patient with intestinal bleeding of unknown etiology, intraoperative endoscopy can be a useful tool to confirm the origin of the hemorrhage.

We observed an extremely rare case of long-term hemosuccus pancreaticus during follow-up for chronic pancreatitis after a Puestow procedure. Intraoperative endoscopy was useful and effective at surgery to confirm the origin of the hemorrhage.

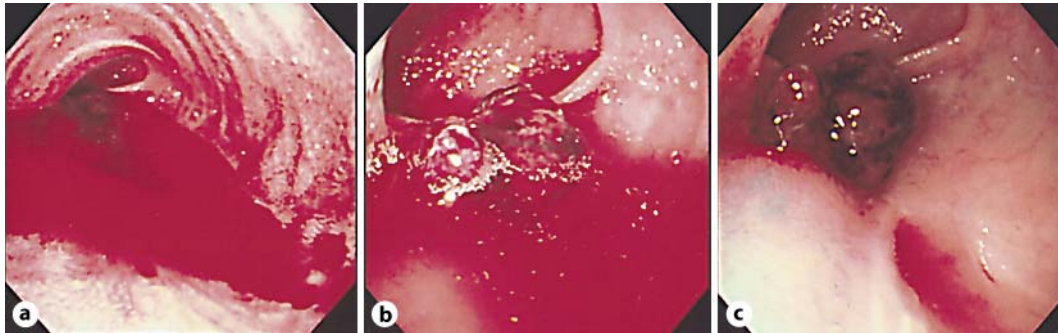


Fig. 1. Intraoperative views on endoscopy inserted from the incised reconstructed jejunum. **a** Active massive bleeding was observed at the site of the side-to-side pancreaticojejunostomy. **b** After suction of the blood, the bleeding point was gradually defined. **c** The bleeding point became clear as coagulation clots in the pancreatic duct epithelium were observed.

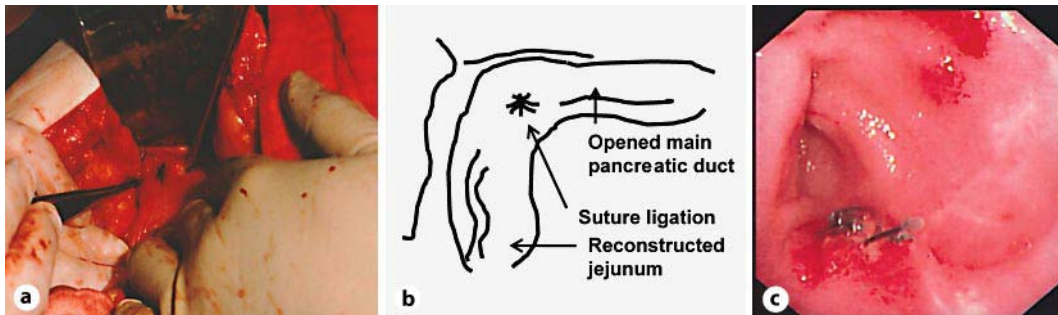


Fig. 2. **a** Intraoperative view of the opened side-to-side pancreaticojejunostomy. A ligated suture could be seen. **b** Illustration of the view in **a**. **c** Endoscopic view of the hemostasis site of the pancreaticojejunostomy after one month. The endoscope was inserted from a percutaneous jejunostomy.

References

- 1 Lower WE, Farrel JJ: Aneurism of the splenic artery. Report of a case and review of literature. *Arch Surg* 1931;23:182–190.
- 2 Sandblom P: Gastrointestinal hemorrhage through the pancreatic duct. *Ann Surg* 1970;171:61–66.
- 3 Lermite E, Regenet N, Tuech JJ, Pessaix P, Meurette G, Bridoux V, Aubé C, Arnaud JP: Diagnosis and treatment of hemosuccus pancreaticus. *Pancreas* 2007;34:229–232.
- 4 Puestow CB, Gillesby WJ: Retrograde surgical drainage of pancreas for chronic relapsing pancreatitis. *Arch Surg* 1958;76:898–904.
- 5 Peroux JL, Arput JP, Saints-Paul MC, Dumas R, Hastier P, Caroli FX, Benchimol D, Delmont JP: Wirsungorrhagie compliquant une pancréatite chronique associée à une tumeur neuro-endocrine du pancréas. *Gastroenterol Clin Biol* 1994;18:1142–1145.
- 6 Menu JA, Fernandez-Cebrian JM, Alvarez-Baleriola I, Barrasa A, Morales V, Carda P: Hemosuccus pancreaticus in a heterotopic jejuna pancreas. *Hepatogastroenterology* 1999;46:177–179.
- 7 Vasquez-Iglesias JL, Durana JA, Yanez J, Rodriguez H, Garcia-Vallejo L, Arnal F: Santorinirrhage: hemosuccus pancreaticus in pancreas divisum. *Am J Gastroenterol* 1988;83:876–878.

- 8 Kuruma S, Kamisawa T, Tu Y, Egawa N, Tsuruta K, Tonooka A, Funata N: Hemosuccus pancreaticus due to intraductal papillary-mucinous carcinoma of the pancreas. *Clin J Gastroenterol* 2009;1:27–29.
- 9 Nezuka H, Yabushita K, Houjou S, Inokuchi M, Fujita H, Yamamoto S, Kaji M, Maeda K, Konishi K: A case of chronic pancreatitis with hemosuccus pancreaticus caused by a microrupture of the splenic artery with arteriosclerosing into the main pancreatic duct. *Jpn J Gastroenterol Surg* 2004;37:1883–1887.
- 10 Mandel SR, Jaques PF, Sanofsky S, Maruro MA: Nonoperative management of peripancreatic arterial aneurysms. A 10-year experience. *Ann Surg* 1987;205:126–128.
- 11 Baker KS, Tisnado J, Cho SR, Beachley MC: Splanchnic artery aneurysms and pseudoaneurysms: transcatheter embolization. *Radiology* 1987;163:135–139.
- 12 Benz CA, Jakob P, Jakobs R, Riemann JF: Hemosuccus pancreaticus – a rare cause of gastrointestinal bleeding: diagnosis and interventional radiological therapy. *Endoscopy* 2000;32:428–431.
- 13 Suzuki T, Hatori T, Imaizumi T, Harada N, Fukuda A, Kamikozuru H, Yazawa T, Noguchi T, Takasaki K: Two cases of hemosuccus pancreaticus in which hemostasis was achieved by transcatheter arterial embolization. *J Hepatobiliary Pancreat Surg* 2003;10:450–454.
- 14 Toyoki Y, Hakamada K, Narumi S, Nara M, Ishido K, Sasaki M: Hemosuccus pancreaticus: problems and pitfalls in diagnosis and treatment. *World J Gastroenterol* 2008;14:2776–2779.