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CASE REPORT

Hemosuccus pancreaticus: Problems and pitfalls in diagnosis and treatment

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Abstract

Hemosuccus pancreaticus is a rare cause of intermittent upper gastrointestinal bleeding. We report two cases of hemosuccus pancreaticus with multiple episodes of upper gastrointestinal bleeding. The causes of hemorrhage were rupture of pseudoaneurysm of the splenic artery and bleeding from the wall of pancreatic pseudocyst. Interventional radiology is the first modality for early diagnosis and possible treatment of hemosuccus pancreaticus. When angiography shows no abnormal findings or interventional radiological therapy can not be successful, surgery should be considered without delay. Our patients herein underwent surgery without recurrence or sequelae. Intraoperative ultrasonography and pancreatoscopy were helpful modalities for confirming the source of hemorrhage and determining the cutting line of the pancreas. When we encounter intermittent upper gastrointestinal bleeding with an obscure source, hemosuccus pancreaticus should be included in differential diagnoses especially in patients with chronic pancreatitis, which would lead to a prompt and proper treatment.

Key words: Hemosuccus pancreaticus; Gastrointestinal bleeding; Interventional radiology; Intraoperative sonography; Intraoperative pancreatoscopy

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INTRODUCTION

Hemorrhage from the papilla of Vater *via* the pancreatic duct, known as hemosuccus pancreaticus, is a rare cause of intermittent upper gastrointestinal bleeding. This condition was first reported in 1931 by Lower and Farrell who mentioned bleeding from an aneurysm of the splenic artery[1]. The expression "hemosuccus pancreaticus" was named by Sandblom in 1970[2]. Until now, reports on hemosuccus pancreaticus have been quite limited. Difficulties in determining the location of bleeding sometimes cause delay of treatment and critical condition of patients.

We herein report two cases of hemosuccus pancreaticus and discuss problems and pitfalls for managing this disease.

CASE REPORTS

Case 1

A 75-year-old woman had been followed up for epigastric discomfort and anemia for 3 years, but no abnormality had been elucidated by either upper gastrointestinal series or endoscopic examinations. She developed sudden hematoemesis and was emergently admitted to a referral hospital. Upper gastrointestinal endoscopy revealed fresh bleeding from the papilla of Vater (Figure 1A), and she was transferred to our institute. A CT scan showed a 2.0 cm × 1.8 cm cystic mass at the tail of the pancreas without remarkable findings of chronic pancreatitis (Figure 1B). Angiography identified aneurysms at the distal portion of the splenic artery and the right hepatic artery (Figure 1C). The patient came down with a pre-shock condition with continuous bleeding, and a diagnosis of the rupture of aneurysm of the splenic artery and/or the right hepatic artery was confirmed emergently. Intraoperative ultrasonography revealed a 2.0 cm × 2.0 cm low echoic mass at the body of the pancreas suspected as a hematoma (Figure 1D). Distal pancreatectomy and splenectomy were performed for the rupture of aneurysm of the splenic artery. The pancreas was diffusely hard and compatible with chronic pancreatitis. It seemed that this might be caused by obstruction of main pancreatic duct due to blood from hematoma. Pathological examination confirmed peripancreatic hematoma and pseudoaneurysm

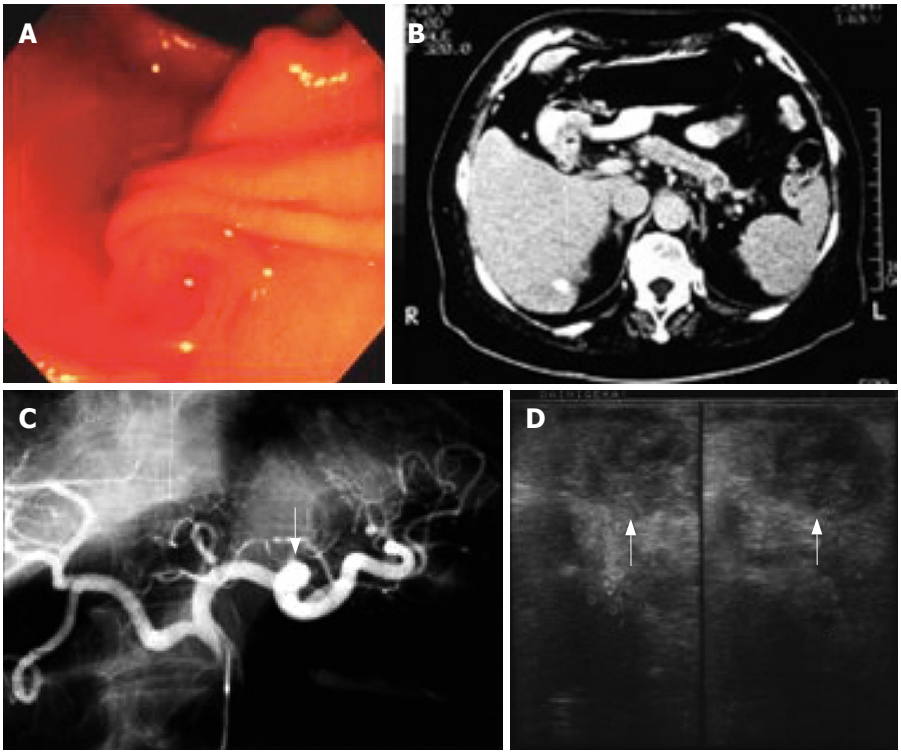

Figure 1 Case 1. Pseudoaneurysm of splenic artery communicating with pancreatic duct. **A**: Endoscopy reveals bleeding from papilla of Vater; **B**: CT scan shows a cystic mass at tail of pancreas; **C**: Angiography identifies aneurysms at distal portion of splenic artery and right hepatic artery (arrow); **D**: Operative ultrasonography demonstrates a low echoic mass at body of pancreas suspected as a hematoma (arrow).

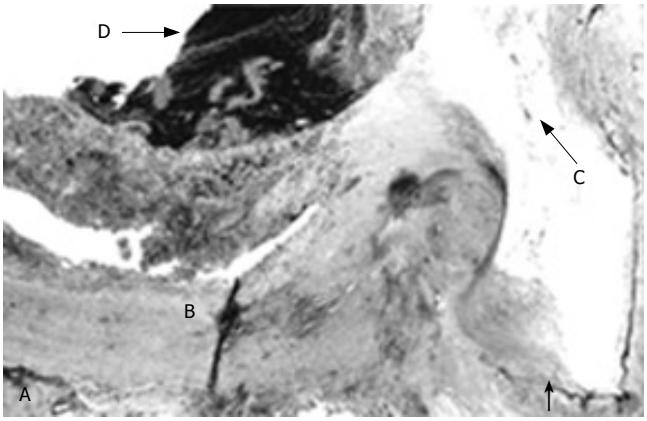

Figure 2 Histology of Case 1. A: Pancreatic parenchyma; B: Wall of pseudoaneurysm of splenic artery; C: Orifice of pseudoaneurysm into pancreatic duct; D: Hematoma.

of the splenic artery communicated with the pancreatic duct: hemosuccus pancreaticus (Figure 2). Postoperative course was uneventful without recurrence.

Case 2

A 44-year-old man was hospitalized at a referral hospital developing tarry stool with severe anemia. Upon the first upper gastrointestinal endoscopy, a small amount of fresh blood was observed in the duodenum, but the source of bleeding was not identified. Colorectal endoscopy, angiography (Figure 3A) and scintigraphy failed to detect the bleeding source. A CT scan revealed multiple calcifications at the whole pancreas and dilatation of main pancreatic duct compatible with chronic pancreatitis and pancreatolithiasis (Figure 3B). Endoscopic retrograde pancreatography and magnetic resonance cholangiopancreatography revealed a dilatated branch of pancreatic duct at the head of the pancreas (Figure 3C). Finally, bleeding from the papilla of Vater was seen by the upper gastrointestinal endoscopy (Figure 3D). He was transferred to our hospital and was electively explored with a diagnosis of hemosuccus pancreaticus and alcoholic chronic pancreatitis. The pancreas was macroscopically compatible with chronic pancreatitis and the bleeding source remained unclear because the stricture of the main pancreatic duct at the head of the pancreas prevented the endoscopic examination *via* the papilla of Vater. The pancreas was then divided right above the portal vein, and the bleeding source was identified at the tail of the pancreas by endoscopic examination (Figure 4A and B). Distal pancreatectomy and splenectomy were performed. Pathological examination demonstrated a pseudocyst filled with hematoma at the tail of the pancreas (Figure 4C).

DISCUSSION

Hemosuccus pancreaticus (HP), a rare cause of upper gastrointestinal bleeding from the papilla of Vater *via* the pancreatic duct, is most commonly caused by the rupture of aneurysm of the splenic artery associated with acute or chronic pancreatitis. Pseudoaneurysm of the hepatic, gastroduodenal or pancreaticoduodenal artery have also been reported as sources of bleeding[3-5]. Other uncommon causes are pancreatolithiasis and pseudocyst of the pancreas[6,7]. Our two patients demonstrated different pathogenetic mechanisms of HP: (1) a rupture of splenic arterial pseudoaneurysm communicating to the pancreatic duct (case 1), and (2) a communication between the peripancreatic artery and pancreatic pseudocyst (case 2).

It is difficult to make HP diagnosis because of intermittent hemorrhage from a source that is not

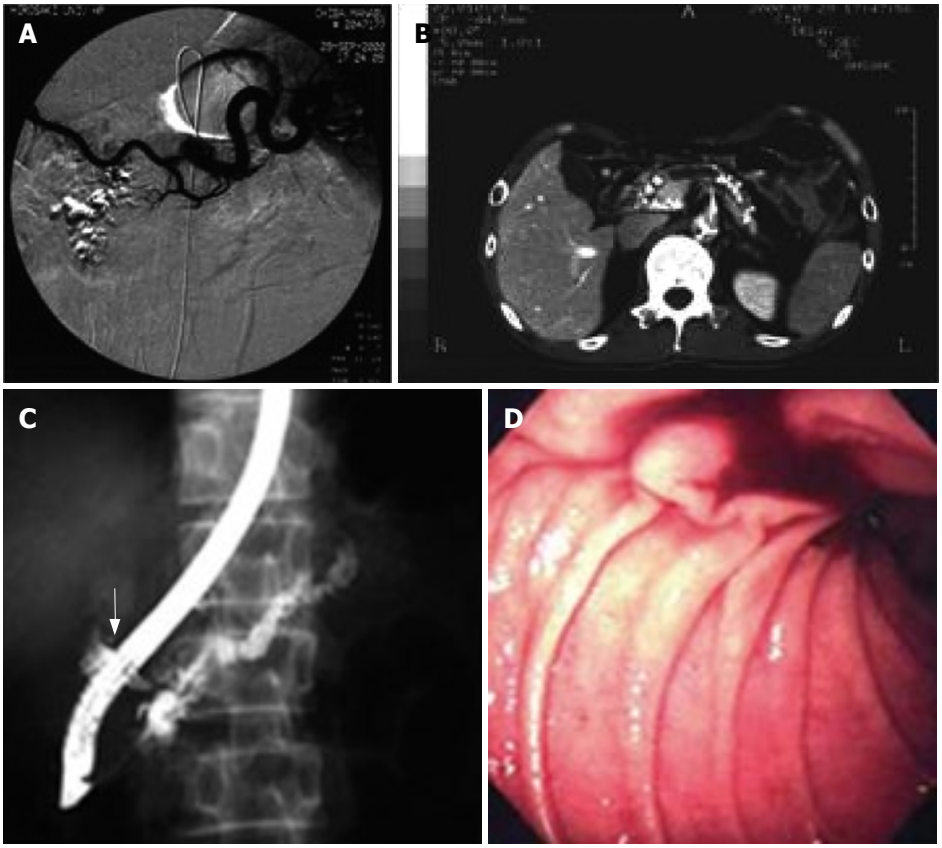

Figure 3 Case 2. Pseudoaneurysm in pancreatic pseudocyst. **A**: Angiography failed to detect a bleeding point; **B**: CT scan shows multiple calcifications at the whole pancreas and dilatation of main pancreatic duct; **C**: Endoscopic retrograde pancreatography displays a dilatated branch of pancreatic duct at head of pancreas (arrow); **D**: Endoscopy reveals bleeding from papilla of Vater.

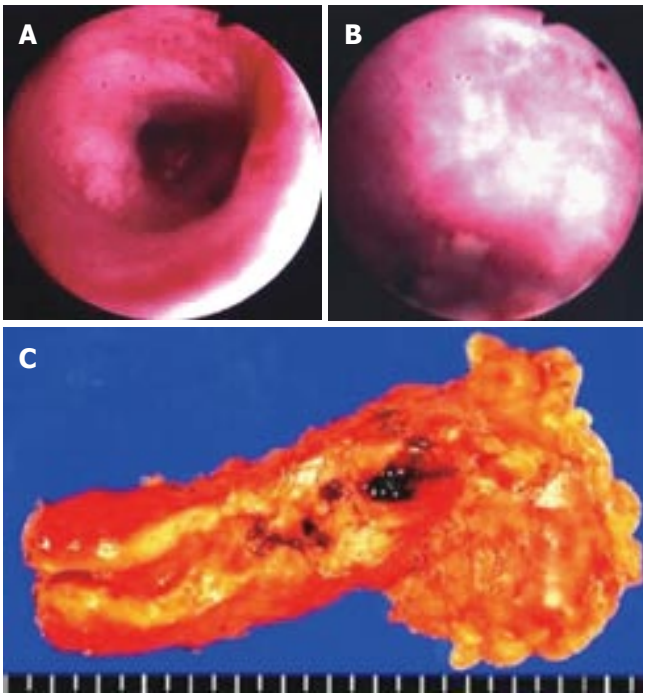

Figure 4 Case 2. Intraoperative pancreatoscopy and specimen. **A**: Pancreatic head by pancreatoscopy; **B**: Pancreatic tail by pancreatoscopy, and bleeding source was seen; **C**: Bleeding source at pancreatic tail.

readily accessible by endoscopy. Moreover, some patients underwent operations elsewhere of questionable benefit before establishment of the correct diagnosis[8]. Therefore, if patients present with obscure source of repeated upper gastrointestinal bleeding, especially underlying chronic pancreatitis, repeated examinations and careful observations should be performed for the diagnosis of these conditions and HP should be included in differential diagnosis[9]. Koizumi *et al*[10] reported that MRI successfully identified the fistula and bleeding. However, MRI was not helpful for the diagnosis of HP in our cases, and reasons remain unknown.

The management for HP should be aimed to eradicate the source of bleeding completely. There are two choices for the treatment of HP: (1) surgery (e.g. resection of the pancreas head or tail), and (2) interventional radiological therapy[11-14]. Most HP cases can receive angiography. If the source of hemorrhage is found by arteriography, interventional radiological therapy should be done following this examination. Recently, Benz *et al*[13] reported the interventional radiological therapy of HP by implantation of an uncoated metal Palmaz stent across the aneurysmal segment of splenic artery. This interventional radiological treatment may be useful for the rupture of arterial pseudoaneurysm to the pancreatic duct so as to prevent emergency operation. However, surgical treatment is required when angiography shows no abnormal findings and interventional radiological therapy is not successful. For the patient with HP who has a pancreatic disease such as pancreatic pseudocyst, surgical treatment may be appropriate. However, it is very difficult to confirm the source of bleeding and determine the cutting line of pancreas. Therefore, intraoperative sonography and pancreatoscopy should be performed to confirm the origin of hemorrhage. They have also been frequently used during hepatobiliary and pancreas surgery. There has been no report about intraoperative ultrasonography

and pancreatoscopy in this disease. Case 1 in our report had two aneurysms of splenic artery and right hepatic artery. Preoperative angiography did not reveal fistula either between aneurysm and pancreatic duct or between aneurysm and bile duct. On intraoperative ultrasonography, no abnormality in hepatobiliary system was seen, but a hematoma could be seen in the pancreatic body. Finally, the diagnosis of a rupture of aneurysm of splenic artery was established. Case 2 in our study was diagnosed with bleeding from the pseudocyst at the pancreatic head by ERCP and MRCP. However, on intraoperative ultrasonography and pancreatoscopy, the bleeding point turned out to be a pseudocyst at the pancreatic tail. Intraoperative pancreatoscopy was also useful in finding the origin of bleeding.

In summary, we had experienced two cases of HP. Repeated examinations and careful observations should be performed to find the obscure source of repeated upper gastrointestinal bleeding and HP should be included in the differential diagnosis. Interventional radiological therapy should be tried at first for HP. Only when angiography shows no abnormal findings and interventional radiological therapy is not successful, surgical treatment is considered. Intraoperative ultrasonography and pancreatoscopy are often performed at surgery to confirm the origin of hemorrhage.

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