CASE REPORT

Hemosuccus pancreaticus: Problems and pitfalls in diagnosis and treatment

Yoshikazu Toyoki, Kenichi Hakamada, Shunji Narumi, Masaki Nara, Keinosuke Ishido, Mutsuo Sasaki

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...
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 cholangio-
pancreatography 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
readily accessible by endoscopy. Moreover, some patients underwent operations elsewhere of questionable benefit before establishment of the correct diagnosis\(^\text{[8]}\). 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\(^\text{[9]}\). Koizumi \textit{et al}\(^\text{[10]}\) 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\(^\text{[11-14]}\). 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 \textit{et al}\(^\text{[13]}\) 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.

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 dilated 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.
and pancreateoscopy 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 pancreateoscopy, the bleeding point turned out to be a pseudocyst at the pancreatic tail. Intraoperative pancreateoscopy 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 pancreateoscopy are often performed at surgery to confirm the origin of hemorrhage.

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