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Case Report

Hemosuccus Pancreaticus in a Patient with Celiac Trunk Aneurysm

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Celiac artery aneurysm associated with hemosuccus pancreaticus is extremely rare. We herein present a 67-year-old man with a large celiac artery aneurysm presenting with intermittent massive gastrointestinal bleeding due to connection of aneurysm with a blood-filled pancreatic duct who was successfully operated. To the best of our knowledge, this is the first report of a case of hemosuccus pancreaticus due to celiac artery trunk aneurysm.

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Introduction

Hemosuccus pancreaticus or wirsungorrhagia, first defined by Sandblom[1] in 1970, is the least common cause of upper gastrointestinal (GI) bleeding in which blood loss occurs into the GI tract through the ampulla of Vater usually due to a communication between one of the branches of visceral artery and Wirsung duct and is similar to clinical syndrome of hemobilia. The first case was described by Lower and Farrell[2] in 1931 which was found to be due to splenic artery aneurysm with communication to the Wirsung duct. The disease is uncommon and up to now about 100 cases have been reported in the literature, 80% of cases associated with chronic pancreatitis with or without pancreatic pseudocyst.[3,4] The etiology of this disease in majority of cases is the pseudoaneurysm of a visceral artery secondary to enzymatic destruction of the arterial wall during pancreatitis.[5] The single most common cause of hemosuccus pancreaticus is rupture of a pseudoaneurysm of the splenic artery associated with pancreatitis.

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Case Report

A 67-year-old man was referred to our hospital for evaluation of acute intermittent massive GI bleeding of obscure origin. Three years earlier, he had his first episode of epigastric pain lasting for one to two hours followed by melena and weakness, which lasted for at least one week. He experienced one such episode every three months until six weeks ago when he developed melena with episodes of hematemesis for which he was admitted in another hospital and received transfusion of five units of packed red blood cells. He was finally transferred to our center for consideration of capsule endoscopy. All the bleeding episodes were preceded by a moderate crampy upper abdominal pain.

He had no other significant medical history and was taking no medications. He denied consumption of illicit substances or alcohol. He had undergone multiple upper endoscopic and colonoscopic examinations in addition to small bowel follow through, red blood cell radionuclide scan, and ultrasonographic studies all failing to demonstrate the site of bleeding. The only positive finding in the colon was a 1-cm polyp at the hepatic flexure and one small polyp in the descending colon both of which were removed.

During one upper endoscopy, fresh blood was seen in the stomach and duodenum. Physical examination was notable for pallor and a soft abdomen with normoactive bowel sounds. Rectal examination revealed black stool. On admission, the hemoglobin was 6.5 g/dL with otherwise unremarkable laboratory values. His hemoglobin value decreased from 13.5 g/dL to 5.6 g/dL during episodes of bleeding and he received repeated blood transfusion during the last three years. After admission to our hospital, he underwent capsule endoscopy which revealed fresh blood in the upper duodenum. A double balloon enteroscopic study was stopped prematurely because of active bleeding seemingly from the ampulla of Vater. The level of serum amylase was 187 IU/L (normal, 110 IU/L).

Urgent upper gastrointestinal (GI) endoscopy and an abdominal plain radiogram were normal. During admission in our hospital, he developed an episode of rebleeding with hematemesis. Upper endoscopy with side view duodenoscope revealed active bleeding coming out of both major and minor papillae (Figure 1). He underwent a spiral CT scan of the abdomen (Figure 2) followed by digital celiac and mesenteric angiography (Figure 3), which revealed a celiac artery aneurysm. Surgical exploration was undertaken, and the aneurysm appeared to be within the pancreatic body (Figure 4) surrounded by a dense, partly-inflamed tissue impossible to be dissected off. Instead of attempts to get access to the origin of celiac artery which was felt to be both difficult and risky, the aorta was freed from above the celiac

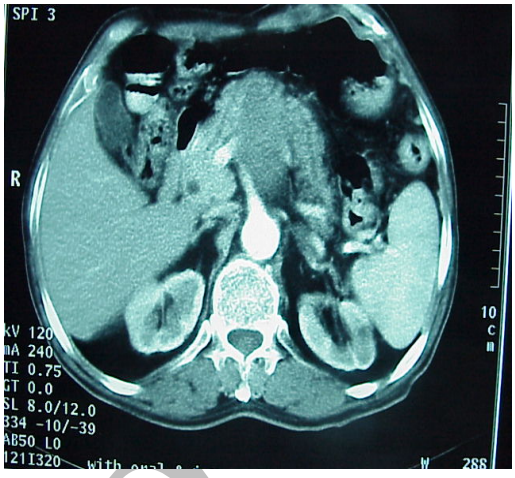

Figure 2. CT-scan with intravenous contrast revealing a low-density mass in posterior to the pancreatic body with extravasations of blood inside it.

artery and below the pancreas. Under systemic heparinization, the aorta was cross- clamped above and below the aneurysm and the aneurysm wall was opened anteriorly. Organized and fresh clots were removed followed by active back bleeding which was controlled by finger pressure from inside the aneurysm cavity. The entrance and exit orifices were sutured, ligated and the rest of the pathology was left in place with no further manipulations. The postoperative course was uneventful and during a two-month follow-up, he remained asymptomatic with no abdominal pain and no further bleeding.

Discussion

This patient was found to have celiac trunk true

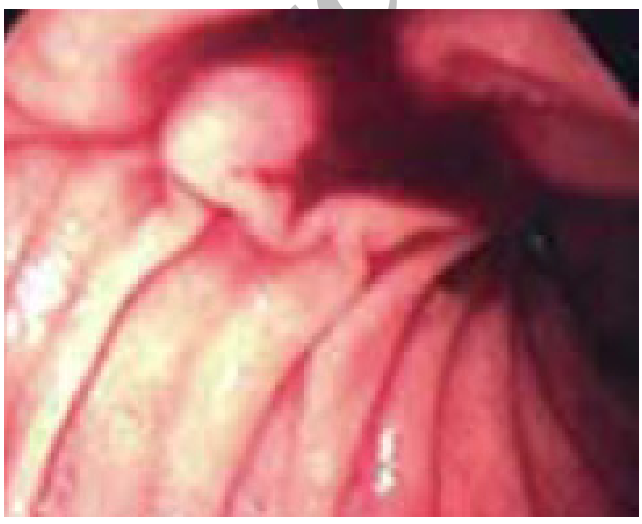

Figure 1. Blood coming out of the ampulla of Vater during side view duodenoscopy.

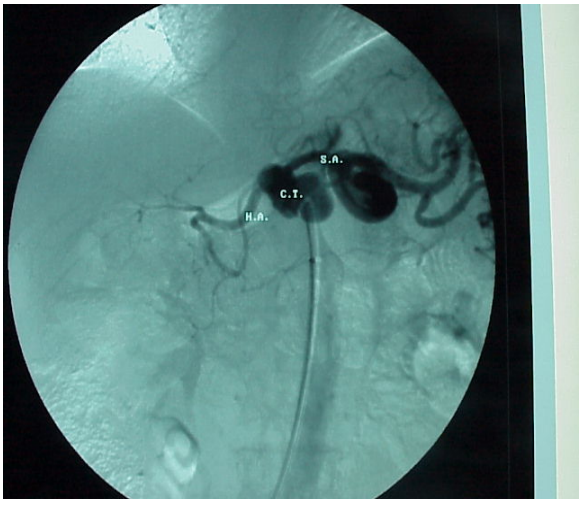

Figure 3. Digital angiography showing a true celiac artery trunk aneurysm with no involvement of the hepatic or splenic artery.

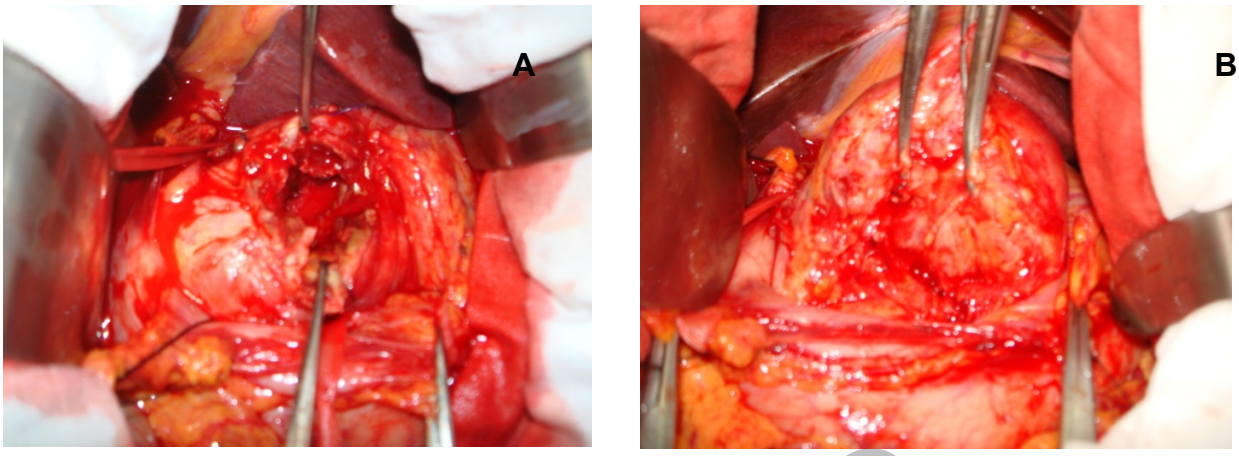

Figure 4 (A and B). True celiac trunk aneurysm within the body of pancreas during operation.

aneurysm with involvement of the pancreas presenting with hemosuccus pancreaticus without evidence of chronic pancreatitis. This is the first case of hemosuccus pancreaticus in association with celiac trunk true aneurysm. In Table 1 we have summarized the etiology for hemosuccus pancreaticus as reported in the literature. The bleeding is arterial in origin except for very rare occasions after rupture of larger veins into a pseudocyst.[6,7] There is a recent case report of hemosuccus pancreaticus caused by spontaneous celiac trunk dissection,[8] but we could not find any case of celiac trunk true aneurysm with involvement of the pancreas presenting with hemosuccus pancreaticus in the literature up to now. Hemosuccus pancreaticus is a disease predominantly occurring in men (M to F ratio= 7:1), with a mean age of onset at about 50 years. Bleeding, although arterial in most instances, is not severe enough to cause hemodynamic instability.[5] The intermittent nature of the bleeding is also very specific and results from formation of the clot in the main pancreatic duct. Other clinical findings, which may occur are: jaundice secondary to blood clot formation in the common channel, vomiting, weight loss, and palpable pulsating epigastric mass with a systolic thrill in the event of aneurysm. Iron-deficiency anemia is frequent but liver function tests are normal except for abnormality in the event of biliary obstruction.[6] Serum amylase is normal except for episodes of acute pancreatitis. Endoscopic retrograde cholangiopancreatography (ERCP) is not recommended for confirmation of diagnosis because CT scan or digital angiography can easily localize the disease and will obviate the need for ERCP and its potential complications.[6]

Table 1. Etiology of hemosuccus pancreaticus.

Pancreatic diseases (80% of all cases)
Chronic pancreatitis
Pseudocyst eroding and communicating with pericyst artery
Intraductal stone or dilatation eroding and producing vascular ulceration
Pseudoaneurysm in visceral artery secondary to chronic pancreatitis
Neuroendocrine tumor of pancreas
Ductal adenocarcinoma of pancreas
Acute pancreatitis
Ectopic pancreas
Pancreas divisum
Post-ERCP
Vascular anomaly in visceral arteries (20%)
True aneurysm of visceral arteries
Pseudoaneurysm of visceral arteries (nonpancreatic origin)
Dissection of visceral arteries

ERCP= endoscopic retrograde cholangiopancreatography

The diagnosis was delayed for three years and the patient underwent repeated endoscopic studies without diagnosis. The key to the diagnosis was the intermittent nature of symptoms preceding the episode of bleeding and the finding of blood coming out of the ampulla of Vater during endoscopy. CT scan, angiography, and ultrasound have all been successful in the identification of pseudoaneurysms and are acceptable diagnostic modalities. Both surgery and endovascular embolization have traditionally been proposed for treatment. No evidence-based guidelines exist regarding the optimal treatment modality as limited data is available. Interventional radiology is the gold standard for early diagnosis and possible treatment of hemosuccus pancreaticus. When interventional radiologic therapy is not available or cannot be successful, surgery should be considered

without delay.[5,6] The surgical procedure of choice in case of splenic artery pseudoaneurysm is distal pancreatectomy with splenectomy. Proximal and distal ligation of the aneurysmal vessel, with oversewing of the ductal communication is the procedure of choice in case of true aneurysm but can also be used in cases of pseudoaneurysm with chronic pancreatitis as a temporizing measure along with drainage of pseudocyst, especially when other surgical approach is not feasible.[4] If conservative transarterial approach was selected in a patient with chronic pancreatitis the remaining diseased pancreas adjacent to previously injured artery may be the source of re-occurrence of arterial injury and bleeding.[4–6]

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