

A Case of Recurrent Infection Caused by a Pancreaticoduodenal Fistula Associated with a Pancreatic Arteriovenous Malformation

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Although arteriovenous malformations (AVM) occur frequently in digestive organs, pancreatic AVM is rare. The clinical symptoms of pancreatic AVM are variable and include gastrointestinal bleeding, abdominal pain, jaundice, portal hypertension, pancreatitis, and duodenal ulcer. However, choledochoduodenal or pancreaticoduodenal fistulas complicated with ascending infection and pancreatitis is extremely rare. Herein, we report a case of pancreaticoduodenal fistula associated with a pancreatic AVM that induced recurrent anemia and ascending infection. (**Gut Liver 2011;5:391-394**)

Key Words: Pancreatic arteriovenous malformation; Pancreaticoduodenal fistula; Infection

INTRODUCTION

Arteriovenous malformation (AVM) probably occurs from loss of the regulatory sphincteric function of the arteriolar-capillary junction which results in overflow of the arterial blood into the capillaries and venules, producing an arteriovenous shunt.¹ Pancreatic AVM is a rare disease which can be complicated with abdominal pain, gastrointestinal bleeding, portal hypertension, and duodenal ulcer.² However, formation of the pancreaticoduodenal fistula by pancreatic AVM is extremely rare. Herein, we report a case of pancreaticoduodenal fistula associated with pancreatic AVM which induced recurrent anemia and ascending infection.

CASE REPORT

A 58-year-old man was admitted to Chonnam National University Hospital with recurrent fever and epigastric pain for 2 months. Two years ago, the patient visited for exertional

dyspnea and he was diagnosed as iron deficiency anemia with pancreatic arteriovenous malformation communicating gastroduodenal artery and pancreaticoduodenal artery with superior mesenteric vein (Fig. 1). Since then he experienced recurrent anemia and he took oral iron preparations occasionally. One month ago, he visited again for fever and epigastric pain. The body temperature was 38.2°C. Laboratory investigation revealed leukocyte 12,900/mm³ (neutrophil 79.5%), hemoglobin 14.1 g/dL, platelet 239,000/mm³. The blood chemistry were protein 6.1 g/dL, albumin 3.2 g/dL, AST 127 U/L, ALT 96 U/L, alkaline phosphatase 145 U/L, total bilirubin 1.1 mg/dL, gamma glutamyl transpeptidase 190 U/L, amylase 85 U/L, lipase 34 U/L, CRP 9.1 mg/dL, prothrombin time 11.8/92.1/1.05 sec/%/INR.

Despite he underwent thorough examinations including computerized tomography (CT) of chest and abdomen, esophagogastroduodenoscopy and colonoscopy, there was no definitive cause of fever. He recovered after 1 week of antibiotics treatment. He had no history of alcohol ingestion.

On admission, the patient appeared acutely ill and he complained tenderness in epigastrium. The body temperature was 36.5°C. Laboratory investigation revealed leukocyte 9,900/mm³ (neutrophil 75.5%), hemoglobin 12.2 g/dL, platelet 337,000/mm³. The blood chemistry were protein 6.8 g/dL, albumin 3.8 g/dL, AST 16 U/L, ALT 22 U/L, alkaline phosphatase 156 U/L, total bilirubin 0.5 mg/dL, gamma glutamyl transpeptidase 94 U/L, amylase 159 U/L, lipase 34 U/L, CRP 2.0 mg/dL, prothrombin time 11.5/96/1.03 sec/%/INR and the urinary analysis was within normal limit.

The chest X-ray was within normal limit. Abdominal CT showed abnormally dilated vessels which abutted with descending duodenum and communicated with gastroduodenal artery and superior mesenteric vein. The head of pancreas was focally enlarged, soft tissue was infiltrated in the peripancreatic fat

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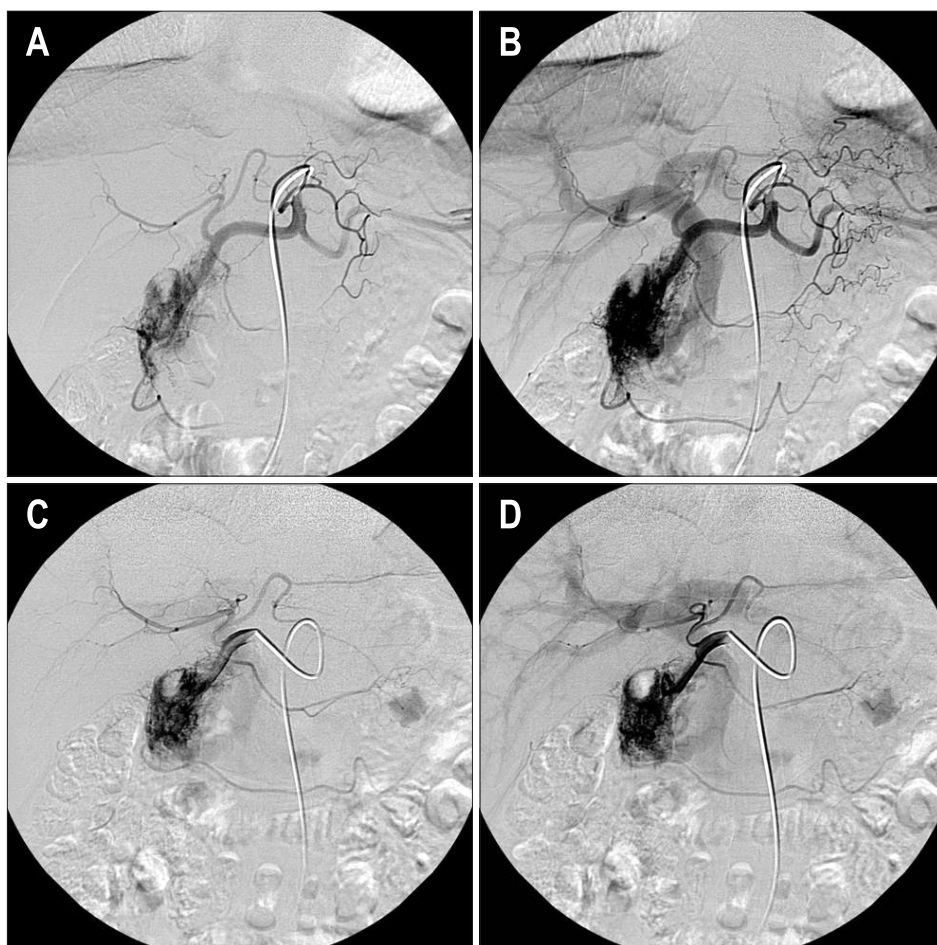


Fig. 1. Angiography. (A) Common hepatic arteriograms demonstrate an abnormal leash of vessels arising from the gastroduodenal artery. (B) Early venous filling of the superior mesenteric vein and portal vein can be seen. (C) Superior mesenteric arteriograms demonstrate an abnormal leash of vessels arising from the superior pancreaticoduodenal artery. (D) Early venous filling of the superior mesenteric vein and portal vein can be seen.

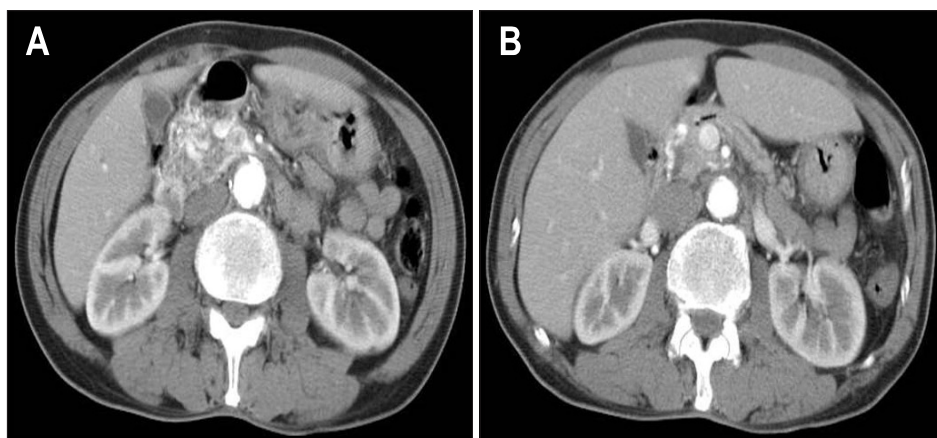


Fig. 2. Abdominal computerized tomogram. (A) Abnormally dilated, conglomerated vascular structures in the pancreatic head abutting the descending duodenum are seen. (B) The pancreatic parenchyma is atrophied except at the pancreatic head, and there are air pockets in the dilated pancreatic duct.

plane, and air densities in the main pancreatic duct were detected (Fig. 2). In the suspicion of pancreaticoduodenal fistula, endoscopic retrograde cholangiopancreatography was undertaken. A fistula hole was detected in the descending duodenum proximal to minor papilla. When the radiocontrast dye was injected through the fistula, the dilated pancreatic duct was visualized (Fig. 3) and the cholangiogram via ampulla of Vater was normal. Review of the past abdominal CT films revealed air densities in the main pancreatic duct also.

On hospital day 5, he complained abdominal pain and the body temperature was 39.5°C. The leukocyte was 12,900/mm³ (neutrophil 91.7%) and *Citrobacter freundii* was cultured from peripheral blood. On hospital day 8, the fever and abdominal pain was relieved with antibiotics.

We recommended surgical therapy for the prevention of portal hypertension and recurrent ascending infection. But, he refused surgery and other conservative treatment including angiographic embolization or transjugular intrahepatic portosys-

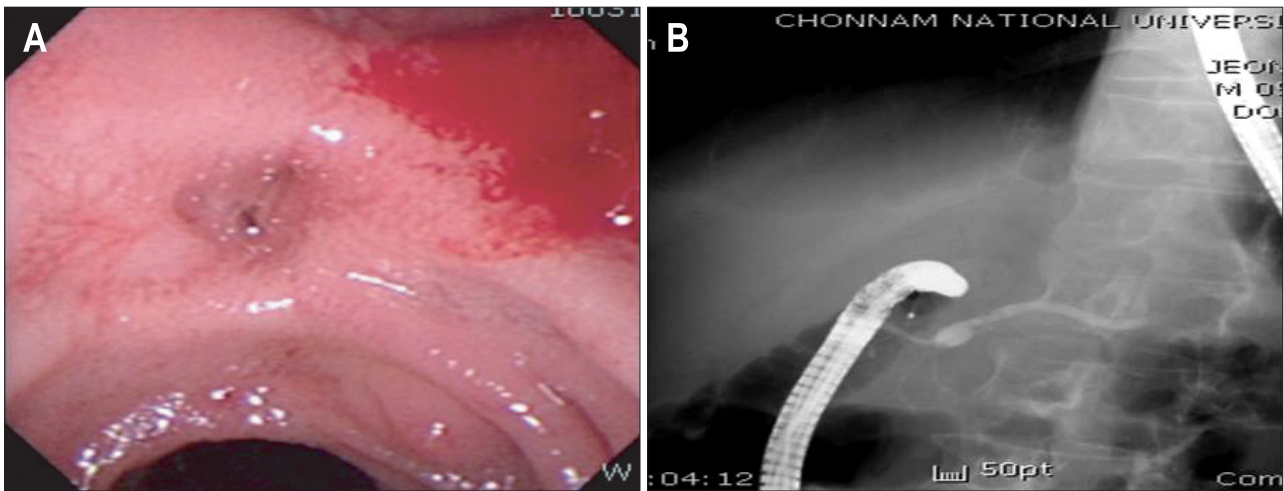


Fig. 3. Endoscopic retrograde cholangiopancreatography. (A) The mucosa of the duodenum and of the fistula located at the descending duodenum proximal to the minor papilla are edematous and easily friable. Around the fistula tract, bleeding telangiectasias can be seen. (B) When the radiocontrast dye is injected through the fistula, the dilated pancreatic duct is visualized.

temic shunt. And after that, he was lost to follow-up.

DISCUSSION

Although AVM can be occurred frequently in digestive organ such as cecum, ascending colon, jejunum, and ileum, pancreatic AVM is rare³ and reported first by Halpern *et al.*⁴ Doppler ultrasonography and CT can be helpful, angiography has played the most important role in the diagnosis and planning the therapy of pancreatic AVM. The angiographic findings of pancreatic AVM include 1) dilated and tortuous feeding arteries, 2) racemose intrapancreatic vascular network followed by a transient dense pancreatic strain, 3) early venous filling into the portal vein, and 4) early disappearance of the pancreatic strain.⁵ The vessels most commonly affected by a pancreatic AVM are the splenic artery (42%), gastroduodenal artery (22%), and small pancreatic arteries (25%).⁶

The clinical symptoms of pancreatic AVM were variable including gastrointestinal bleeding, abdominal pain, jaundice, portal hypertension, pancreatitis, and duodenal ulcer.^{2,7} Gastrointestinal bleeding can be occurred by following mechanisms, first, direct intestinal bleeding from intestinal mucosa, second, bleeding from pancreatic duct, and third, bleeding from esophagogastric varices associated with portal hypertension.⁸ In the present case, although there was no evidence of massive gastrointestinal bleeding, chronic occult blood loss might have occurred from telangiectasia associated with pancreatic AVM. Duodenal ulcer or duodenitis appears to be associated with regional ischemia caused by the diseased mucosa.⁹ Recurrent inflammation of duodenum and pancreas might have induced the fistula tract between pancreatic duct and duodenum. However, choledochoduodenal or pancreaticoduodenal fistula associated with pancreatic AVM is extremely rare, and according to the English literature, only 2 cases have been reported^{10,11} and

so far, there was no report about recurrent ascending infection caused by pancreaticoduodenal fistula associated with pancreatic AVM. *Citrobacter freundii* is gram negative enteric bacilli which can be found in human intestine and infects urinary and biliary tract. In the present case, there was no evidence of infection focus other than pancreaticoduodenal fistula. Invasion of *Citrobacter freundii* via pancreaticoduodenal fistula might have induced pancreatitis and bacteremia. In the present case, the patient's pancreatic parenchyma was atrophied and the pancreatic duct was dilated. Chronic ischemia caused by pancreatic AVM could induce chronic pancreatitis and chronic pancreatitis was acutely exacerbated at each time of ascending infection by pancreaticoduodenal fistula. It is known that pancreatitis associated with pancreatic AVM can be occurred by bleeding from AVM into the pancreatic duct and ischemia of the pancreas.⁷ Ascending infection via pancreaticoduodenal fistula should be considered as a cause of pancreatitis.

Complete cure is achieved by total resection of the affected organ, or at least the involved portion.¹² Once after portal hypertension developed, it cannot be corrected even after surgical resection, so early diagnosis and proper treatment is important.⁵ In case of inoperable conditions, transcatheter arterial embolization,¹² radiation therapy,¹³ and transjugular intrahepatic portosystemic shunt¹⁴ seems to be the alternative treatment for complications of pancreatic AVM.

In summary, pancreatic AVM is a rare disease, but can induce life threatening complications. Herein, we report a case of pancreaticoduodenal fistula associated with pancreatic AVM which induced recurrent anemia and ascending infection.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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