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Case Report

Endovascular treatment of a pancreatic artery pseudoaneurysm with arterioportal fistula in chronic pancreatitis: A case report ☆,☆☆

Miki Yoshida, MD[a,], Kensaku Mori, MD[b], Sakiko Shigeta, MD[c], Masaomi Nagase, MD[c], Kazunori Ishige, MD[c], Kuniaki Fukuda, MD[c], Masato Endou, MD[d], Tsukasa Saida, MD[e], Takahito Nakajima, MD[e]*

[a] *Department of Diagnostic and Interventional Radiology, University of Tsukuba Hospital, 2-1-1 Amakubo, Tsukuba, Ibaraki 305-8576, Japan*
[b] *Department of Radiology, The Cancer Institute Hospital of Japanese Foundation for Cancer Research, 3-8-31 Ariake, Koto, Tokyo 135-8550, Japan*
[c] *Department of Gastroenterology and Hepatology, Tsukuba Gakuen Hospital, 2573-1, Kamiyokoba, Tsukuba, Ibaraki 305-0854, Japan*
[d] *Department of gastroenterology, University of Tsukuba Hospital, 2-1-1 Amakubo, Tsukuba, Ibaraki 305-8576, Japan*
[e] *Department of Diagnostic and Interventional Radiology, Institute of Medicine, University of Tsukuba, 1-1-1 Tennodai, Tsukuba, Ibaraki 305-8575, Japan*

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ABSTRACT

Arterioportal fistula (APF) combined with a visceral artery pseudoaneurysm is an exceptionally rare and critical vascular disorder of the abdominal viscera, with pseudoaneurysm rupture being potentially fatal and severe APF leading to portal hypertension, both of which necessitate immediate intervention. An 87-year-old woman with a history of pancreatitis presented with upper abdomen and back pain. Laboratory tests revealed elevated amylase levels and severe anemia. A computed tomography (CT) scan showed a large dorsal pancreatic artery (DPA) pseudoaneurysm with a fistula to the main portal vein. Given her advanced age, surgery was deemed high-risk, and endovascular treatment was selected. Transcatheter arterial embolization was successfully performed using coils to embolize the

Abbreviations: APF, arterioportal fistula; DPA, dorsal pancreatic artery; CT, computed tomography; TAE, transcatheter arterial embolization; SMA, superior mesenteric artery; IPDA, inferior pancreaticoduodenal artery; SMV, superior mesenteric vein; ASPDA, anterior superior pancreaticoduodenal artery; GDA, gastroduodenal artery.

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* Corresponding author.
E-mail address: yoshida.miki.ce@ms.hosp.tsukuba.ac.jp (M. Yoshida).

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DPA pseudoaneurysm. A follow-up CT 1 week postprocedure confirmed the absence of a pseudoaneurysm and no further progression of anemia.

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Introduction

Chronic pancreatitis is an ongoing fibroinflammatory disease of the pancreas characterized by irreversible damage to the pancreatic parenchyma and ductal system [1]. During its progression, leaking digestive enzymes may erode nearby vessels and form pseudoaneurysms, which can become a fatal complication if they rupture [1,2]. Arterioportal fistula (APF) is a direct communication between the arterial circulation and the portal system [3]. APF can be classified as congenital or acquired, intrahepatic or extrahepatic [4]. The inflow can originate from any of the splanchnic arteries, with the most common being the hepatic artery (hepatoportal fistula, 65%), and reports of extrahepatic APF are few [3,4]. APFs are a rare cause of presinusoidal portal hypertension [4]. Pseudoaneurysms can occur as a complication of chronic pancreatitis, but we cannot find any reports of this forming an APF. We report a case of a postpancreatitis a pseudoaneurysm and large fistula between the dorsal pancreatic artery (DPA) and main portal vein, which was successfully treated with a transcatheter arterial embolization (TAE).

Case description

The patient was an 87-year-old woman with a history of surgery for ovarian cancer. She had been noted to have a 6 mm cystic lesion in the pancreatic head on contrast-enhanced computed tomography (CT) 3 years prior. The lesion was monitored over time with CT and magnetic resonance imaging (MRI), showing progressive enlargement. She had no history of hospitalization for pancreatitis. She presented to her physician with upper abdomen and back pain. Physical examination revealed minimal abdominal abnormalities. Laboratory tests showed anemia (hemoglobin, 8.2 g/dL) and elevated pancreatic enzyme levels (amylase, 530 U/L). Contrast-enhanced CT revealed a hemorrhage within a pancreatic head cyst. There was no active bleeding at that time, the initial management included treatment for pancreatitis and blood transfusion, after which she was discharged. Three months later, her symptoms recurred, and she was brought to the emergency department because of immobility. Laboratory tests revealed severe anemia (hemoglobin, 3.2 g/dL) and increased pancreatic enzyme levels (amylase, 553 U/L). Imaging findings revealed a 3.7 cm saccular aneurysm of the DPA with an APF on contrast-enhanced CT (Fig. 1). Owing to her advanced age, surgery such as pancreaticoduodenectomy was considered burdensome and high-risk, making it difficult to perform. Therefore, less invasive endovascular treatment was chosen. The patient was transferred to our hospital, and the procedure was performed the following day.

We punctured the right common femoral artery and inserted a 4F sheath (Medikit, Tokyo, Japan). Diagnostic superior mesenteric artery (SMA) and celiac artery arteriographies were performed using a 4F JA3 catheter (Medikit, Tokyo, Japan), which revealed a large DPA pseudoaneurysm with APF (Fig. 2). SMA angiography showed early delineation of the known aneurysm through a collateral pathway via the inferior pancreaticoduodenal artery. The intrahepatic portal vein was delineated through a fistula between the aneurysm and main trunk of the portal vein. The superior mesenteric vein (SMV) was delineated caudal to the aneurysm but did not extend into the liver. Instead, the intrahepatic portal vein was drawn from the main portal vein via a collateral path around the pancreatic head and through the fistula. Celiac arteriography depicted the pseudoaneurysm via the DPA from an early stage and the intrahepatic portal vein from the main portal vein via a fistula. We planned to embolize the DPA pseudoaneurysm using a transarterial approach.

We selected the anterior superior pancreaticoduodenal artery via the gastroduodenal artery, using a double coaxial microcatheter system consisting of an outer 2.6F high-flow microcatheter and an inner 1.9 Fr thin microcatheter (Fig. 3). The distal DPA was selected, and digital subtraction angiography was performed. The branches of the DPA visible on the preoperative CT were not visualized. Using the preoperative CT as a reference, the branch was successfully selected with a heat-molded microcatheter, and coil embolization was performed (AZUL soft3D, Terumo, Tokyo, Japan). Additional branches were identified from the proximal side as well, and after performing coil embolization up to the proximal side (AZUL soft3D, Terumo, Tokyo, Japan), the branches were no longer visible.

Next, the proximal DPA originating from the splenic artery was selected using a double coaxial system. To prevent rupture of the pseudoaneurysm, we paid careful attention to avoid coil protrusion into the pseudoaneurysm. Additionally, the short distance to the vessel made coil placement challenging. During the procedure, 1 coil could not be properly deployed and was retrieved, while another coil was damaged and could not be deployed. To facilitate coil placement, the catheter was exchanged from JA3 to JL4, and after advancing the microcatheter with its tip inserted into the DPA, successful coil placement was achieved (AZUL soft3D, Terumo, Tokyo, Japan; Interlock, Boston Scientific, Massachusetts, USA). The final angiogram indicated a complete obliteration of the pseudoaneurysm (Fig. 4). The portal vein was delineated normally from the SMV, and blood flow within the mass through the portal vein was not delineated.

A contrast-enhanced CT performed 1 week after TAE confirmed the absence of the pseudoaneurysm, and there was no

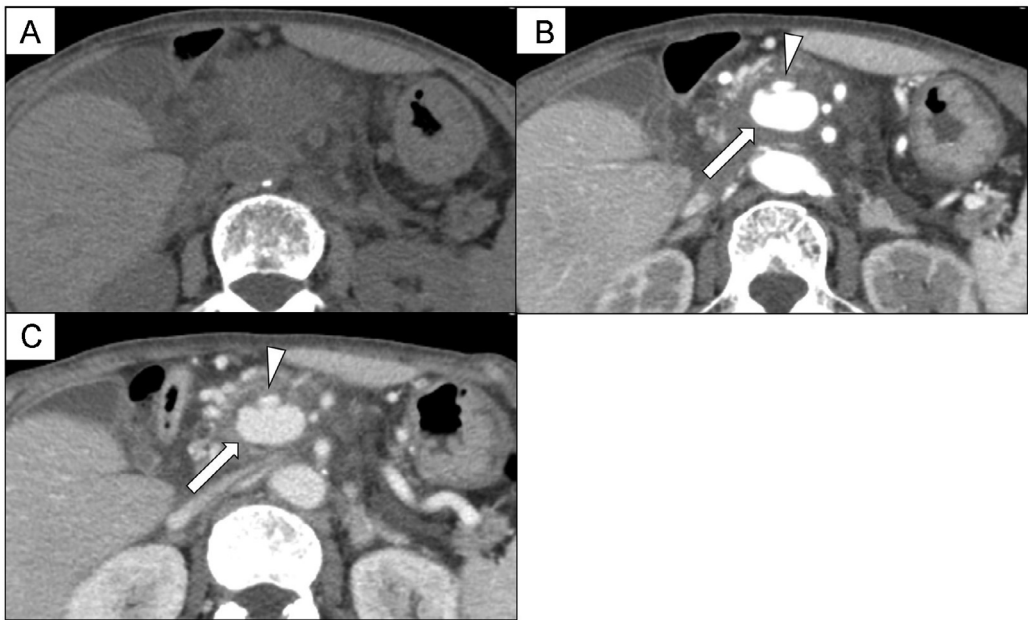

Fig. 1 – (A) NONcontrast-enhanced computed tomography, (B) arterial phase contrast-enhanced computed tomography, (c) portal vein phase contrast-enhanced computed tomography. Contrast-enhanced computed tomography revealed hemorrhage within a pancreatic head cyst. The main portal vein (arrowheads) runs adjacent to the pseudoaneurysm of the dorsal pancreatic artery (arrows), with both structures communicating, diagnosable as arterioportal fistula.

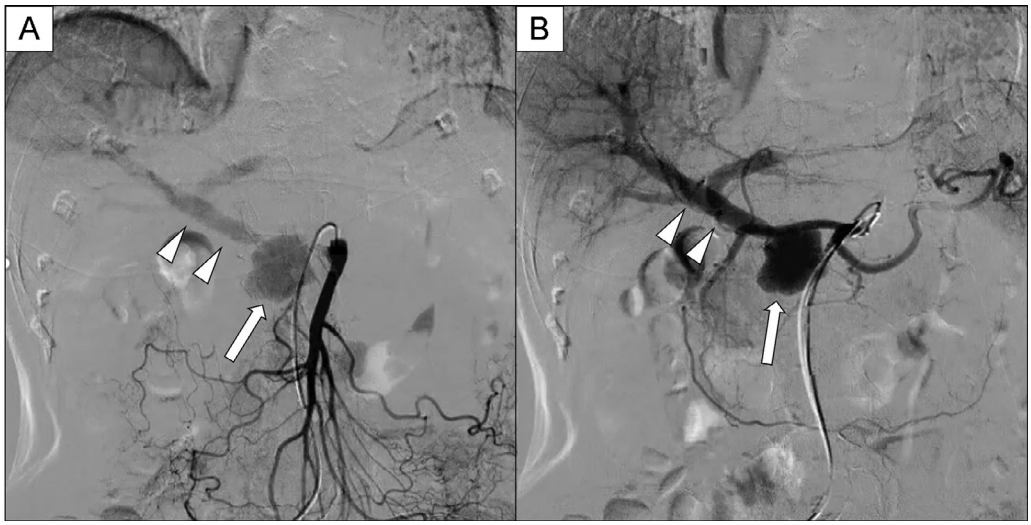

Fig. 2 – (A) the superior mesenteric artery angiography, (B) celiac arteriography. Angiography from the superior mesenteric artery and celiac trunk demonstrate a pseudoaneurysm (A, B; arrows) and early filling of the portal vein (A, B; arrowheads) with an arterioportal fistula.

progression of anemia. The patient's progress was good, and she was transferred to her previous physician for rehabilitation. After discharge, the patient has been regularly attending outpatient follow-ups and remains in good condition without any signs of rebleeding.

Discussion

Chronic pancreatitis is an ongoing fibroinflammatory disease of the pancreas characterized by irreversible damage to the

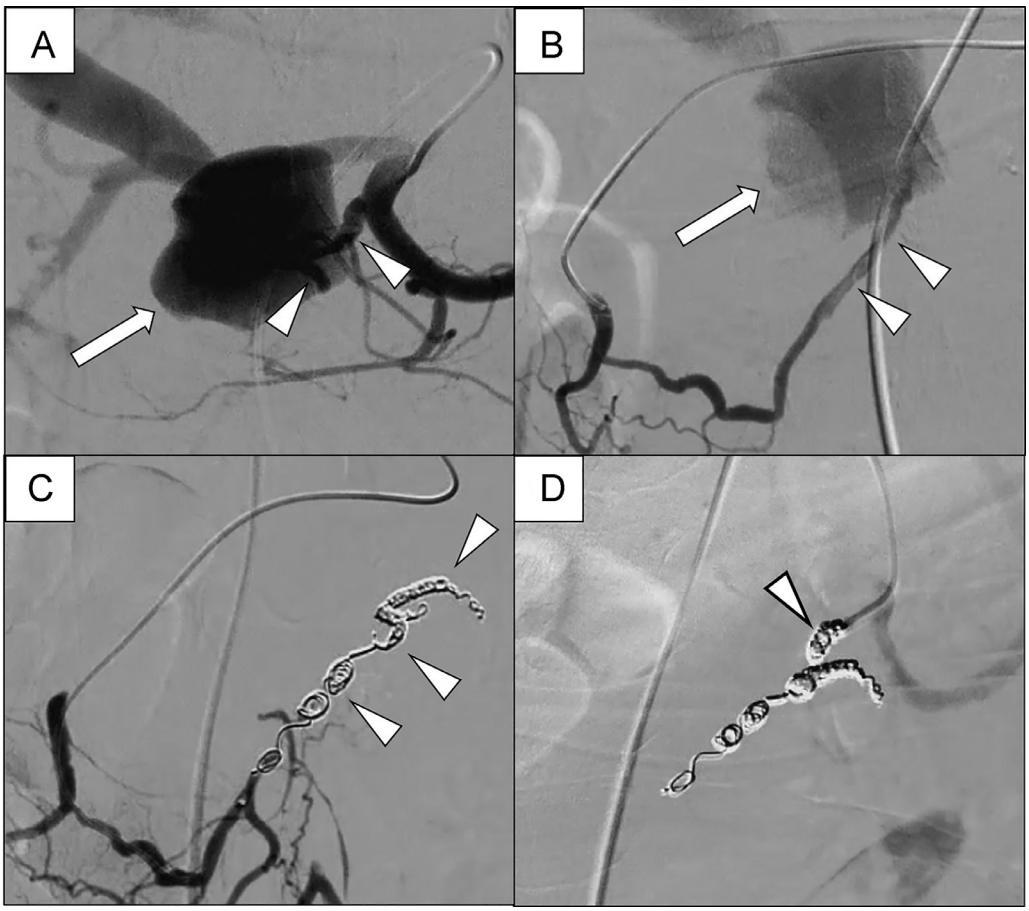

Fig. 3 – The distal and proximal dorsal pancreatic artery embolization using coils. (A) Celiac angiography was performed with a cranial angulation of 14 degrees. The angiography revealed the presence of a pseudoaneurysm (arrow) in the dorsal pancreatic artery (DPA) (arrowheads). (B, C) Distal DPA (arrowheads) is selected via the anterior superior pancreaticoduodenal artery using a double coaxial microcatheter system and embolized using coils (AZUL soft3D, Terumo, Tokyo, Japan). (D) Arrowhead: Coiling proximal DPA (AZUL soft3D, Terumo, Tokyo, Japan; Interlock, Boston Scientific, Massachusetts, USA).

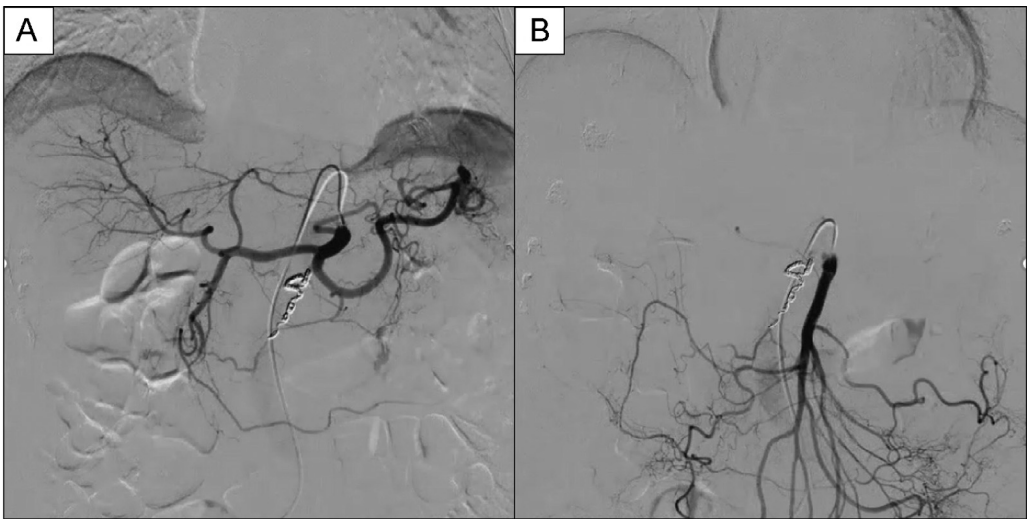

Fig. 4 – (A) celiac angiography, (B) the superior mesenteric artery angiography. Angiography from the superior mesenteric artery and celiac trunk after transcatheter arterial embolization. The pseudoaneurysm disappears with no early filling of the portal vein.

pancreatic parenchyma and ductal system. During its progression, pseudoaneurysms may form because of the erosion of nearby vessels caused by leaking digestive enzymes [1,5]. The reported incidence of pseudoaneurysm formation from pancreatitis varies from 1.3% to 10% [6]. A visceral artery pseudoaneurysm can be a lethal complication of chronic pancreatitis if bleeding occurs [2]. APF is a rare vascular disorder of the abdominal viscera, characterized by arteriovenous communication between the splanchnic arteries and the portal vein or its tributaries [3,7]. APFs are more commonly intrahepatic than extrahepatic, with approximately 15% caused by ruptured splanchnic artery aneurysms [4,8]. They can be acquired or congenital. The most common cause of acquired APF is trauma; other causes include iatrogenic procedures, complications of tumors, and rupture of pseudoaneurysms [3,7,9].

Visceral artery pseudoaneurysms and APF are rare. The patient had elevated amylase levels, a history of previous episodes of pancreatitis, and a pseudocyst on the pancreatic head. The pseudoaneurysm of the DPA that developed in the pseudocyst is believed to have formed due to the erosion of the DPA by leaked digestive enzymes resulting from chronic pancreatitis. Additionally, the micro-rupture of the pseudoaneurysm and repeated erosion by digestive enzymes may have interacted to contribute to the formation of an APF. In this case, contrast-enhanced CT revealed early enhancement of the portal vein in the arterial phase, leading to the diagnosis of APF. Similar findings were observed on digital subtraction angiography of the SMA and celiac artery. To the best of our knowledge, there have been 2 reported cases of fistula of the peripancreatic arteries and the portal vein caused by simultaneous enzyme destruction of a pancreatic pseudocyst [7,10]. Both cases involved pseudoaneurysms of the gastroduodenal artery (GDA). In 1 case, the fistula was occluded using a detachable balloon [10]. In the other case, a balloon catheter was placed in the portal vein and the pseudoaneurysm was embolized using coils and N-butyl cyanoacrylate (NBCA) [7]. Recently, endovascular treatment has become the mainstream approach for managing pseudoaneurysms. Endovascular treatment is also being used for the management of extrahepatic APF, with various methods being employed depending on the case. Reports using stent grafts have also been observed [11,12]. In this case, we were able to treat the patient by completely embolizing the responsible vessel with coils, without the need for a balloon catheter or covered stent. However, depending on the vascular anatomy, different treatment options, such as NBCA or covered stents, should always be considered. Six months after treatment, the patient was hospitalized once for pancreatitis but did not experience anemia, and symptoms improved, allowing for discharge within a week. The patient continues to attend follow-up appointments, and ongoing management of pancreatitis will be necessary based on symptoms and blood tests.

Although chronic pancreatitis causes many vascular complications, simultaneous occurrence of these lesions is extremely rare. Herein, we share our experience with a case of an extrahepatic APF induced by the rupture of DPA pseudoaneurysm and chronic pancreatitis. In this case, we were able to treat the patient by completely embolizing the responsible vessel with coils.

Patient consent

A written consent was obtained from the patient for publication of this case and any accompanying images.

REFERENCES

[1] Brock C, Nielsen LM, Lelic D, Drewes AM. Pathophysiology of chronic pancreatitis. World J Gastroenterol 2013;19:7231–40.
[2] Chiang K-C, Chen T-H, Hsu J-T. Management of chronic pancreatitis complicated with a bleeding pseudoaneurysm. World J Gastroenterol 2014;20:16132–7.
[3] Bonvini RF, Alerci M, Antonucci F, Vandoni RE, Braghetti A, Gertsch P, et al. Extrahepatic arterioportal fistula: three-dimensional reconstruction of computerized tomodensitometric scan for diagnosis and morphologic assessment. Abdom Imaging 2004;29:463–6.
[4] Guzman EA, McCahill LE, Rogers FB. Arterioportal fistulas: introduction of a novel classification with therapeutic implications. J Gastrointest Surg 2006;10:543–50.
[5] Chapa UK, Dutta S, Abhinaya R, Jain A, Kalyanasundaram A, Munuswamy H, et al. Giant celiac artery pseudoaneurysm in a case of chronic pancreatitis: a rare case report with literature review. Vasc Endovascular Surg 2021;55:658–62.
[6] Chapman BM, Bolton JS, Gioe SM, Conway WC. Gastroduodenal artery pseudoaneurysm causing obstructive jaundice. Ochsner J 2021;21:104–7.
[7] Lim SG, Park SE, Nam IC, Choi HC, Won JH, Jo SH, et al. Large gastroduodenal artery pseudoaneurysm, arterioportal fistula and portal vein stenosis in chronic pancreatitis treated using combined transarterial embolization and transportal stenting: a case report. Medicine (Baltimore) 2022;101:e32593.
[8] Waguri N, Azumi R, Osaki A, Sato M, Aiba T, Furukawa K. Ruptured pancreaticoduodenal pseudoaneurysm causing arterioportal fistula: combined transarterial and transportal embolization. CVIR Endovasc 2020;3:72.
[9] Losey AD, Lokken RP, Kolli KP, Kerlan RK, Taylor AG, Kohi MP. Embolization of arterial-portal fistula to treat associated hemobilia after transjugular liver biopsy. Semin Intervent Radiol 2020;37:430–3.
[10] Denys A, Hammel P, De Baere T, Vilgrain V, Bernades P, Roche A, et al. Arterioportal fistula due to a ruptured pancreatic pseudocyst: diagnosis and endovascular treatment. Am J Roentgenol 1998;170:1205–6.
[11] Al-Khayat H, Haider HH, Al-Haddad A, Al-Khayat H, Ginzburg E. Endovascular repair of traumatic superior mesenteric artery to splenic vein fistula. Vasc Endovascular Surg 2007;41:559–63.
[12] Narayanan G, Mohin G, Barbery K, Lamus D, Nanavati K, Yrizarry JM. Endovascular management of superior mesenteric artery pseudoaneurysm and fistula. Cardiovasc Intervent Radiol 2008;31:1239–43.