INTERVENTIONAL RADIOLOGY
Official journal of the Japanese Society of Interventional Radiology and Asia Pacific Society of Cardiovascular and Interventional Radiology

CASE REPORT

Endovascular Treatment of Postpancreatectomy Hemorrhage from the Retroportal Artery with Compression of the Celiac Trunk by the Median Arcuate Ligament: A Case Report

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Abstract:
Retroportal artery is one of the communicating arteries between the hepatic artery and the superior mesenteric artery, but it is often a small artery and usually unrecognized. We report a 60-year-old man that was successfully treated for postpancreatectomy hemorrhage from the retroportal artery with compression of the celiac trunk by the median arcuate ligament. Following the pancreaticoduodenectomy, the bloody discharge was discovered through the drainage catheter. We underwent transcatheter arterial embolization for the bleeding from the retroportal artery associated with a postoperative pancreatic fistula. Additionally, because a stenosis of the common hepatic artery was discovered, we consequently installed a stent-graft on the common hepatic artery to prevent the liver failure due to decreased hepatic blood flow.

Keywords:
postoperative pancreatic fistula, retroportal artery, transarterial embolization, median arcuate ligament

Interventional Radiology 2024; 9(1): 36-40
https://doi.org/10.22575/interventionalradiology.2023-0002
https://ir-journal.jp/

Introduction

The retroportal artery is a small branch of the superior mesenteric artery (SMA), providing an anastomosis with the hepatic artery. This vessel typically supplies the common bile duct and is not usually seen on arteriography, unless it provides a collateral blood flow to the liver [1]. Median arcuate ligament syndrome (MALS) produces an eccentric narrowing due to extrinsic compression, while celiac artery stenosis due to atherosclerosis causes concentric and calcified stenosis. There exists a network of collaterals between the celiac artery and SMA. Narrowing of the celiac artery results in diversion of blood from the SMA through these pathways. Therefore, division of the gastroduodenal artery during pancreaticoduodenectomy can result in liver ischemia [2]. In addition, previous reports described abdominal visceral artery aneurysms associated with MALS. The pancreaticoduodenal artery is the primary site of aneurysm formation in these situations, followed by the middle colic and dorsal pancreatic arteries; however, few reports mention aneurysms in the retroportal artery [3]. We describe a case of bleeding from postoperative pancreatic fistula (POPF) in a patient with MALS treated with embolization of the retroportal artery and stenting to the common hepatic artery to maintain hepatic blood flow.

Case Report

A 60-year-old man underwent drainage for POPF after subtotal stomach-preserving pancreaticoduodenectomy (SSPPD) to treat pancreatic head cancer. A preoperative contrast-enhanced CT (CE-CT) did not show aneurysm of the pancreatic arcade and stenosis of the common hepatic artery. And also, preoperative CE-CT indicated MALS but not retroportal artery from SMA. Coagulated blood was removed from the drainage tube 16 days following surgery. CE-CT showed no pseudoaneurysm; consequently, the patient was maintained under close supervision. However, massive hematemesis occurred the following day. On physical examination, the body temperature was 36.9° C, blood pressure 96/66 mmHg, heart rate 60/min, SpO_2 99% (room air), and breathing rate 18/min. The abdomen was distended with tenderness at the strongest point in the pericardial fossa, though no muscle guarding was observed. Blood tests

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Received: January 27, 2023, Accepted: October 7, 2023

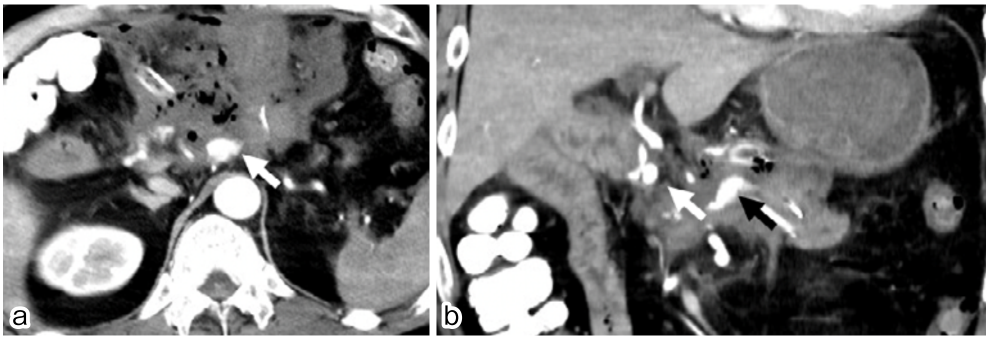

Figure 1. Arterial phase of contrast-enhanced (CE)-CT showing a pseudoaneurysm (white arrow) and extravasation (black arrow) in proximity to an artery running dorsally to the portal vein and connecting the right hepatic artery (RHA) and SMA. a) Axial section. b) Coronal section.

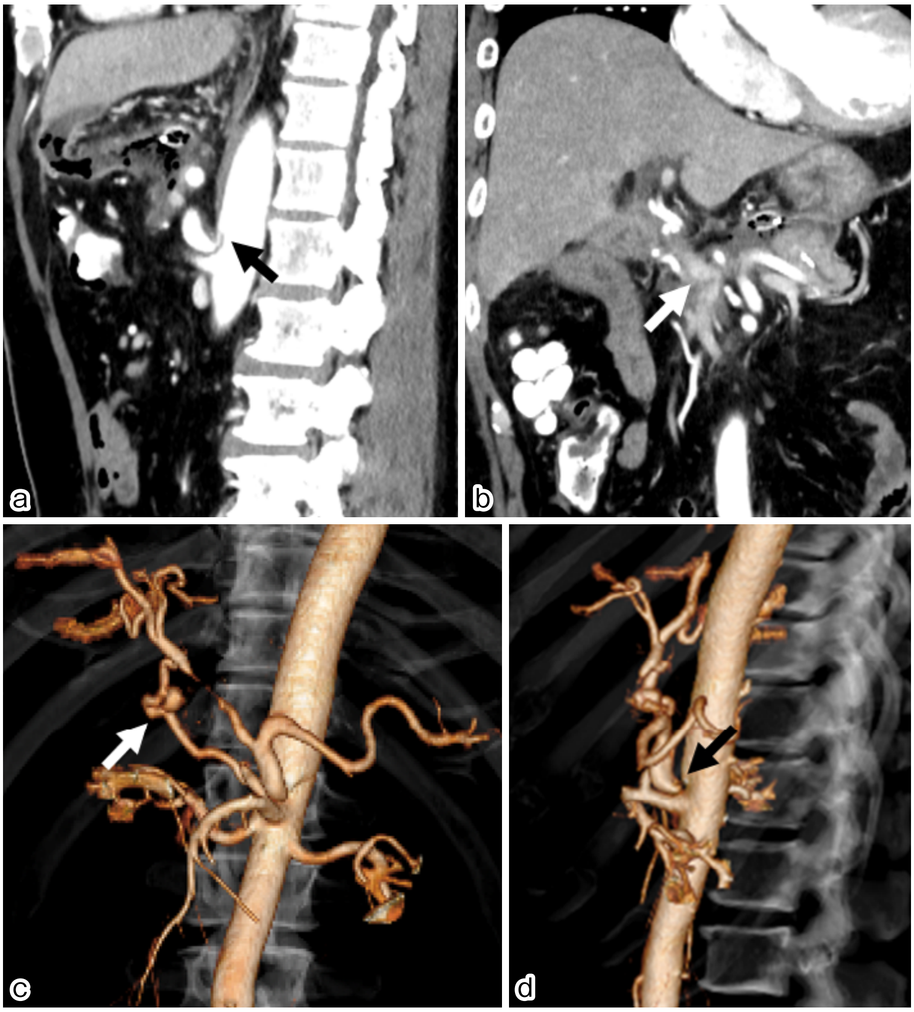

Figure 2. a) Sagittal sections of contrast-enhanced (CE)-CT showing the typical features of median arcuate ligament syndrome (MALS). The celiac axis is compressed by the median arcuate ligament (black arrow).
b) Coronal sections of contrast-enhanced (CE)-CT showing the stenosis of the portal vein caused by POPF (white arrow).
c, d) The 3D images of the entire celiac artery and superior mesenteric artery region including the pseudoaneurysm (PA view and sagittal view). The white arrow indicates the pseudo-an-eurysm (c), and the black arrow indicates the stenosis of the celiac artery (d).

revealed a hemoglobin concentration of 9.6 g/dL and white blood cell count of 10,450/μL. Laboratory examinations demonstrated elevated aspartate aminotransferase (AST = 129 IU/L), alanine aminotransferase (ALT = 78 IU/L), and

Figure 3. Intraoperative angiographic images.
a) SMA angiography showing a pseudoaneurysm in the retroportal artery (white arrow). The proper hepatic artery is also visible.
b) In the portal phase of SMA angiography, portal vein stenosis was also found (black arrow).
c) Embolization with a mixture of 25% NBCA and lipiodol under flow control with detachable coils.
d) Hemostasis was confirmed by postprocedural SMA angiography. The blood flow in the proximal side of the retroportal artery was maintained.
e) Celiac angiography showing a reduced caliber of the common hepatic artery (CHA) (black arrow), although the intrahepatic arterial flow seems adequate.

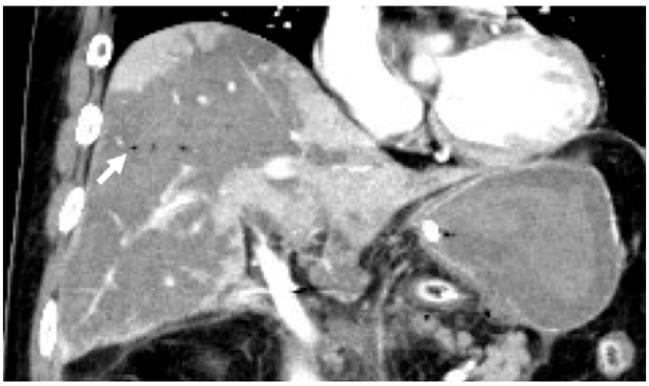

Figure 4. Eight hours after TAE, a contrast-enhanced (CE)-CT showed a large poorly enhanced area in the liver and hepatic portal venous gas (arrow).

C-reactive protein (CRP = 14.4 mg/dL). The unenhanced CT showed a hematoma within the POPF. In the CE-CT, extravasation and a pseudoaneurysm which had been revealed neither on preoperative CT images nor on the CT performed on the day before were noticed in the hematoma (**Fig. 1a** and **1b**). The bleeding artery was identified posteriorly to the portal vein and was connected with the right hepatic artery (RHA) and the SMA. Stenosis of the celiac trunk due to compression by median arcuate ligament and stenosis of the common hepatic artery (CHA) and the portal vein (PV) due to POPF were also detected (**Fig. 2a**-**2d**).

We decided to perform an emergency transcatheter arterial embolization (TAE). We inserted a 4-French (Fr) sheath introducer into the right common femoral artery, and a 4-Fr shepherd hook-type catheter (Medikit, Tokyo, Japan) was advanced into the SMA. SMA angiography revealed dilatation of the retroportal artery, with a pseudoaneurysm on the distal end (**Fig. 3a**). Hepatopetal PV flow was also detected, while PV stenosis was observed in the portal phase (**Fig. 3 b**). We inserted a 2.2-Fr microcatheter (Coiling Support; HI-LEX Medical, Hyogo, Japan) into the retroportal artery antegradely from the SMA. Coil embolization of the distal pseudoaneurysm was performed with two detachable coils (Target XL; Stryker, Kalamazoo, MI, USA; 5 mm × 100 mm) to achieve flow control. Then, the pseudoaneurysm and the proximal side were embolized using 0.8 mL of a mixture of 25% NBCA and lipiodol (**Fig. 3c** and **3d**). Angiography of the celiac artery showed stenosis of the CHA; nevertheless, adequate blood flow was shown in the hepatic artery (**Fig. 3e**).

The patient experienced hematemesis and hypotension 7 hours after previous treatment. An additional CE-CT was performed; no active bleeding was detected, though a large area in the liver was poorly enhanced and hepatic portal ve-

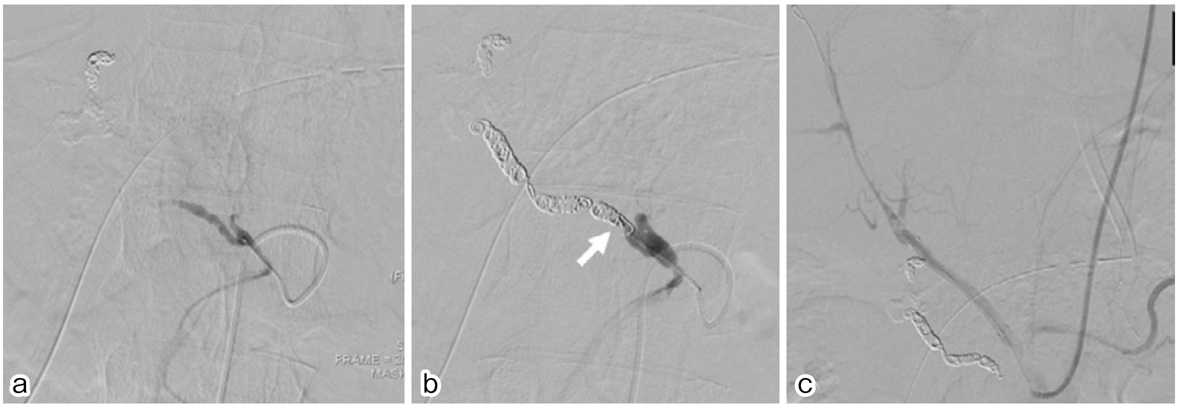

Figure 5. Intraoperative images of the second treatment. a) Selective arteriography of the retroportal artery before additional embolization. b) Additional coil embolization performed on the proximal side of the retroportal artery (arrow). c) The stent-graft insertion repaired the common hepatic artery (CHA) and increased the hepatic blood flow.

nous gas was present (**Fig. 4**). The CHA and the PV were stenosed but patent in that CT same as before. Laboratory examinations revealed elevated levels of ALT (= 309 IU/L), AST (= 439 IU/L), and serum total bilirubin (T-Bil = 1.6 mg/dL). These results led to the diagnosis of hepatic infarction. Additional endovascular treatment was performed to improve the CHA blood flow and achieve hemostasis, if necessary. A 4-Fr sheath introducer was punctured into the left common femoral artery, and the SMA was catheterized using a 4-Fr catheter (Rosch Hepatic; Medikit, Tokyo, Japan). No active bleeding was seen in the superior mesenteric angiography and retroportal angiography (**Fig. 5a**); however, we performed additional coil embolization of the proximal side of the retroportal artery. A 2.2-Fr microcatheter (SIR-ABE, PIOLAX, Yokohama, Japan) was advanced into the retroportal artery through the 4-Fr catheter. Then, we positioned nine pushable coils (Tornado Coil, Cook, Bloomington, IN, USA) with diameters of 3, 4, 5, and 6 mm (**Fig. 5 b**). Since the celiac arteriography showed CHA stenosis, we also placed a stent-graft in this artery. A 7-Fr guiding sheath was injected using the left brachial artery (Destination; Terumo, Tokyo, Japan); then, balloon dilatation of the celiac trunk and CHA was performed using an angioplasty balloon catheter (Sterling; Boston Scientific, Marlborough, MA, USA; 4 mm × 40 mm). Finally, a stent-graft (VIABAHN, W. L. Gore & Associates, Inc., Flagstaff, AZ, USA; 5 mm × 50 mm) was inserted into the CHA. Post-dilatation was also performed using the angioplasty balloon catheter (Sterling; Boston Scientific, Marlborough, MA, USA; 4 mm × 40 mm). Postoperative celiac arteriography showed improvement of the hepatic blood flow (**Fig. 5c**).

Rebleeding was not observed after the endovascular treatment. The increased liver enzyme level is found (AST = 13150 IU/L and ALT = 5552 IU/L) on 1 day and the higher bilirubin level (T-Bil = 25.2 mg/dL) on 14 days after stent-graft placement. The liver enzymes normalized (AST = 24 IU/L, ALT = 23 IU/L) and the bilirubin level decreased (minimum 3.2 mg/dL) in 3 weeks after that, and no progression to liver failure was observed. However, the patient died 71 days following surgery due to a POPF-related infection.

Discussion

The retroportal artery runs dorsally to the PV and pancreatic head and supplies blood to the common bile duct [4]. It is not typically visualized on angiography; however, it is sometimes identified in patients with MALS as an anastomotic branch between the SMA and RHA [5]. Bleeding from the retroportal artery caused by POPF in cases of MALS has not before been reported. In several cases with severe stenosis of the celiac trunk due to compression by median arcuate ligament, the hepatic blood flow is supplied by the SMA via pancreatic arcades. However, in this instance, the postoperative CT did not demonstrate the emergence of pancreatic arcades; in contrast, the dilated retroportal artery providing a collateral blood flow to the liver is visualized. The gastroduodenal artery (GDA) clamp test is usually performed during pancreaticoduodenectomy in patients with MALS to assess whether additional revascularization surgery is required [2]. In this case, the GDA clamp test was negative despite MALS, suggesting a different collateral flow other than pancreatic arcades.

Hemoperitoneum associated with a POPF occurs in 6%-26% of patients after pancreaticoduodenectomy (PD) [6]. It is crucial to monitor the drainage tubes carefully because sentinel bleeding can be seen in 89% of patients 1-2 days before a massive hemorrhage [7]. TAE is one of the effective treatments for bleeding from POPF. However, a POPF after PD may reduce the caliber of the PV, and it has been reported that TAE in these patients can result in liver infarction or abscess. The peripheral arterial stent-graft allowed us to preserve the hepatic blood flow when there was bleeding. There are some reports that especially for the patients with acute bleeding, stent-graft placement should be considered and its patency rates are high in the short term yet decrease as time passes [8-10]. Additionally, most of them stated that covered stent placement was a highly efficient, safe interventional treatment for hemorrhage associated with POPF with positive technical and clinical outcomes, although this case does not involve bleeding from CHA.

In this case, bleeding from the retroportal artery, the main hepatoenteric collateral, probably occurred due to the ligation of the pancreatic arcades that provided an alternative collateral flow, in addition to the POPF involvement. Additionally, TAE of the retroportal artery, associated with the reduced PV and CHA due to POPF, presumably induced liver infarction. In cases of bleeding from the retroportal artery related to POPF, it may be preferable to place a stent-graft at an early stage in case of CHA stenosis; however, long-term evidence is insufficient. Stent-graft placement into the retroportal artery could be a possible procedure at the first time of bleeding retrospectively. However, peripheral arterial stent-grafts were not readily available in our hospital at midnight. And also, the failure to measure hepatic arterial pressure before and after the embolization procedure was a point of reflection in this case. There was no conclusive proof that actual changes in blood flow had occurred.

In summary, we described a case of bleeding from the retroportal artery due to POPF and the hemodynamic change associated with the ligation of the gastroduodenal artery. We managed to achieve hemostasis and preserve the hepatic blood flow; nonetheless, the patient later died because of uncontrolled infection related to POPF. This is the first account of bleeding from the retroportal artery caused by POPF in a patient with MALS.

Conflict of Interest: None

Funding: This study was not supported by any funding.

Author Contribution: Conceptualization, MT, Methodology, MT, Clinical Investigation,TK, MT, RK, AU, and AH, Data curation, TK, MT, RK, AU, and AH, Writing, TK, and MT, Supervision, KI

Informed Consent: Written informed consent was acquired from the patient for the publication of this case report and any accompanying images.

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