

A traumatic hepatic artery pseudoaneurysm and arterioportal fistula, with severe diarrhea as the first symptom

A case report and review of the literature

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Abstract

Rationale: Hepaticarterioportal fistula (APF) is a rare cause of portal hypertension and gastrointestinal hemorrhage, and presents as abnormal communication between the hepatic artery and portal vein. Percutaneous liver biopsy is a main iatrogenic cause of APF. However, non-iatrogenic, abdominal, trauma-related APF is rarely reported.

Patient concerns: A 29-year-old man presenting with severe, watery diarrhea was transferred to our hospital, and his condition was suspected to be acute gastroenteritis because he ate expired food and suffered a penetrating abdominal stab wound 5 years ago. After admission, the patient suffered from hematemesis, hematochezia, ascites, anuria, and kidney failure, and he developed shock.

Diagnoses: The patient was finally diagnosed as a traumatic hepatic artery pseudoaneurysm and APF.

Interventions: This patient was treated with emergency transarterial embolization using coils. Since a secondary feeding vessel was exposed after the first embolization of the main feeding artery, a less-selective embolization was performed again.

Outcomes: During the 6-month follow-up period, the patient remained asymptomatic.

Lessons: A penetrating abdominal stab wound is a rare cause of hepatic APFs, and occasionally leads to portal hypertension, the medical history and physical examination are the most important cornerstones of clinical diagnosis. Interventional radiology is essential for the diagnosis and treatment of an APF.

Abbreviations: APF = arterioportal fistula, BP = blood pressure, CT = computed tomography, DSA = digital subtraction angiography, LPV = left portal vein, MDCT = multidetector computed tomography, TAE = transarterial embolization, THPAD = transient hepatic parenchymal attenuation differences.

Keywords: arterioportal fistula, diarrhea, portal hypertension, pseudoaneurysm, transarterial embolization, traumatic

1. Introduction

Portal hypertension is defined as an elevation of the portal pressure gradient, and it is directly related to the portal blood flow and vascular resistance. Severe portal hypertension causes

symptoms or complications such as diarrhea, gastrointestinal hemorrhage, ascites, and encephalopathy.^[1,2] The causes of portal hypertension can be classified as pre-hepatic (portal vein or splenic thrombosis), intrahepatic (cirrhosis), and posthepatic (Budd–Chiari syndrome).^[1] Hepatic arterioportal fistula (APF) is a rare cause of portal hypertension.^[3] It may be a congenital syndrome, or it is sometimes acquired and develops secondary to cirrhosis^[4]; a hepatic neoplasm^[5]; and trauma, including liver biopsy,^[6] resection,^[7] and transplantation.^[8] We reported the case of a 29-year-old man who presented with portal hypertension and gastrointestinal hemorrhage and was finally diagnosed as having a traumatic hepatic artery pseudoaneurysm and APF. He was successfully treated with transarterial embolization (TAE) twice.

2. Case report

This study was approved by the Ethics Committee of Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology. Written informed consent was obtained from the patient.

A 29-year-old man was transferred to our hospital with complaints of abdominal pain, watery diarrhea, and hematochezia. He experienced severe diarrhea 20 days before admission, after he consumed expired food. Ten days before admission, these symptoms worsened. His stool frequency increased to more than

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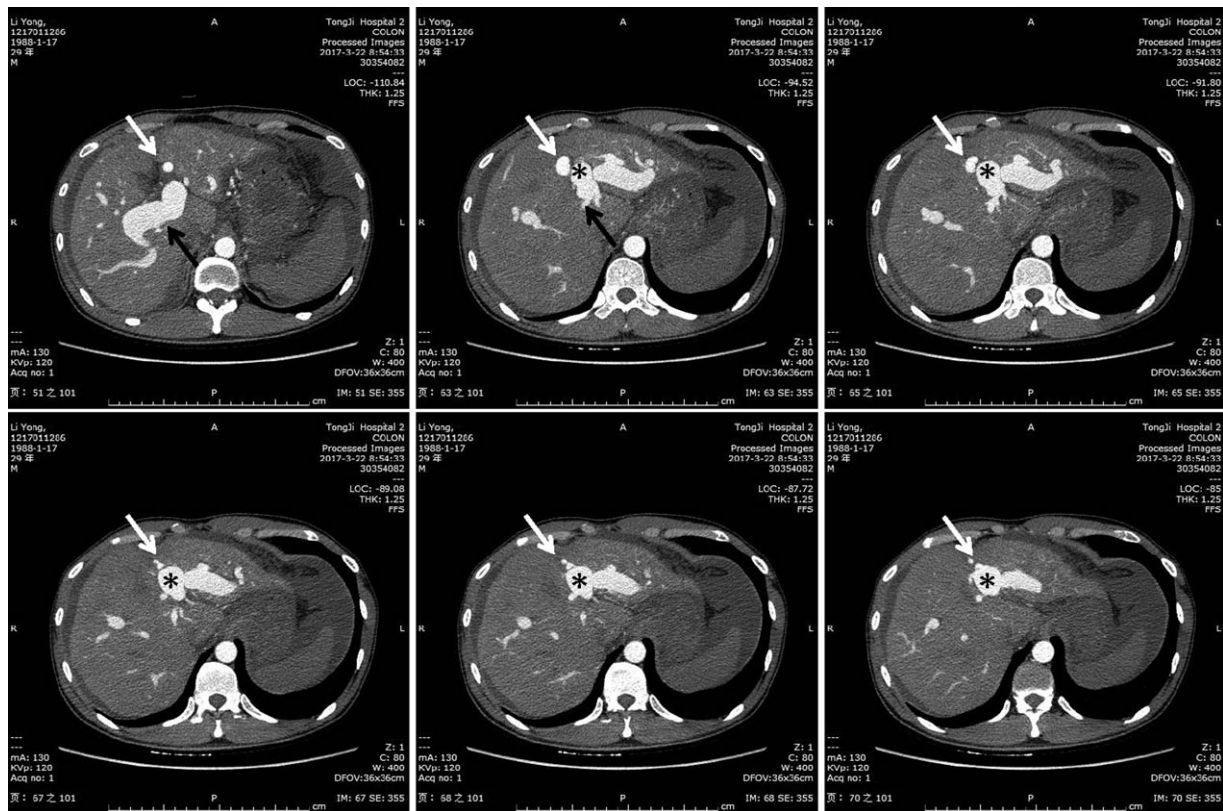


Figure 1. Contrast-enhanced CT shows a typical hepatic artery pseudoaneurysm and APF. The dilated portal vein in the arterial phase is shown (black arrows). The left hepatic artery was enlarged (white arrows) and communicated with the left portal vein. An aneurysmal structure had developed at the site of the APF (black stars). APF=arterioportal fistula, CT=computed tomography.

20 per day, and was accompanied by abdominal pain, nausea, fatigue, oliguria, and hematochezia. He could not tolerate the frequent diarrhea and abdominal pain and attempted to commit suicide. He did not have special medical history other than a penetrating abdominal stab wound resulting in liver injury 5 years ago. The patient consented to undergo surgical repair.

At the time of admission to our hospital, he was not icteric, had a pulse rate of 76 beats/min, and was normotensive [blood pressure (BP) 115/80 mm Hg]. An abdominal examination revealed that a systolic murmur was heard on auscultation over the liver in the right upper quadrant, and there was tenderness in the right quadrant and peri-umbilicus. The remaining physical examination findings were unremarkable. Laboratory findings showed a hemoglobin level of 85 g/L, white blood cell count of $15.64 \times 10^9/L$, and platelet count of $319 \times 10^9/L$. The liver function test results were serum alanine aminotransferase of 36 U/L, serum aspartate aminotransferase of 33 U/L, serum gamma-glutamyltransferase of 159 U/L, serum alkaline phosphatase of 103 U/L, total bilirubin of 17.7 $\mu\text{mol/L}$, direct bilirubin of 8.9 $\mu\text{mol/L}$, and albumin of 39.2 g/L. The kidney function tests showed a blood urea nitrogen level of 22.57 mmol/L, creatinine level of 245 $\mu\text{mol/L}$, uric acid level of 511.3 $\mu\text{mol/L}$, and prothrombin time of 16.7 seconds. A routine stool test showed a red blood cell count of 3 to 4/haptoglobin, white blood cell count of 0/haptoglobin, occult blood positivity in the stool, and an erythrocyte sedimentation rate of 62 mm/H. Alpha-fetoprotein, carcinoembryonic antigen, and carbohydrate antigen-199 were all within the normal range. All viral markers for hepatitis were negative. A T-Spot tuberculosis test for *Mycobacterium tubercu-*

losis was negative. Auto-antibodies (anti-nuclear, anti-mitochondrial, anti-smooth muscle, anti-soluble liver antigen, and anti-mitochondrial antibodies) were also negative.

The initial diagnosis was acute gastroenteritis, until a computerized tomography (CT) scan was performed (Fig. 1). CT with intravenous contrast showed abnormal, early opacity of the portal vein in the arterial phase. The portal vein was markedly dilated, measuring up to 2.4 cm in diameter. The left hepatic artery was enlarged to 1.5 cm and contrast material was pooled in the left anterosuperior area of the liver. A large, left hepatic lobe APF was identified between the left hepatic artery and the left portal vein (LPV). Arterial-phase imaging showed an aneurysmal structure at the site of the APF. All these findings were consistent with a hepatic artery pseudoaneurysm and APF between the LPV branch and left hepatic artery (Fig. 2A and B). Other features, including esophageal and gastric varices, diffuse bleeding of the jejunum, ascites, and gastrointestinal wall thickening, were also found on the CT images (Fig. 2A).

A Doppler ultrasound showed normal liver, intra- and extrahepatic bile ducts, cholecyst, spleen, and pancreas morphology. It also revealed a 2.2×1.6 cm, mildly hyperechoic mass in the left medial segment of the liver, with a well-defined border and slightly irregular contour, which was considered a scar after trauma. The main portal vein was dilated, with a diameter of 2.3 cm. Meanwhile, the LPV branch and left hepatic artery were obviously enlarged near the scar, with diameters of 2.4 and 1.0 cm, respectively (Fig. 2C). Contrast-enhanced ultrasonography that was performed using an intravenous injection of 1-mL SonoVue (Bracco, Italy) revealed that the portal vein synchronously appeared

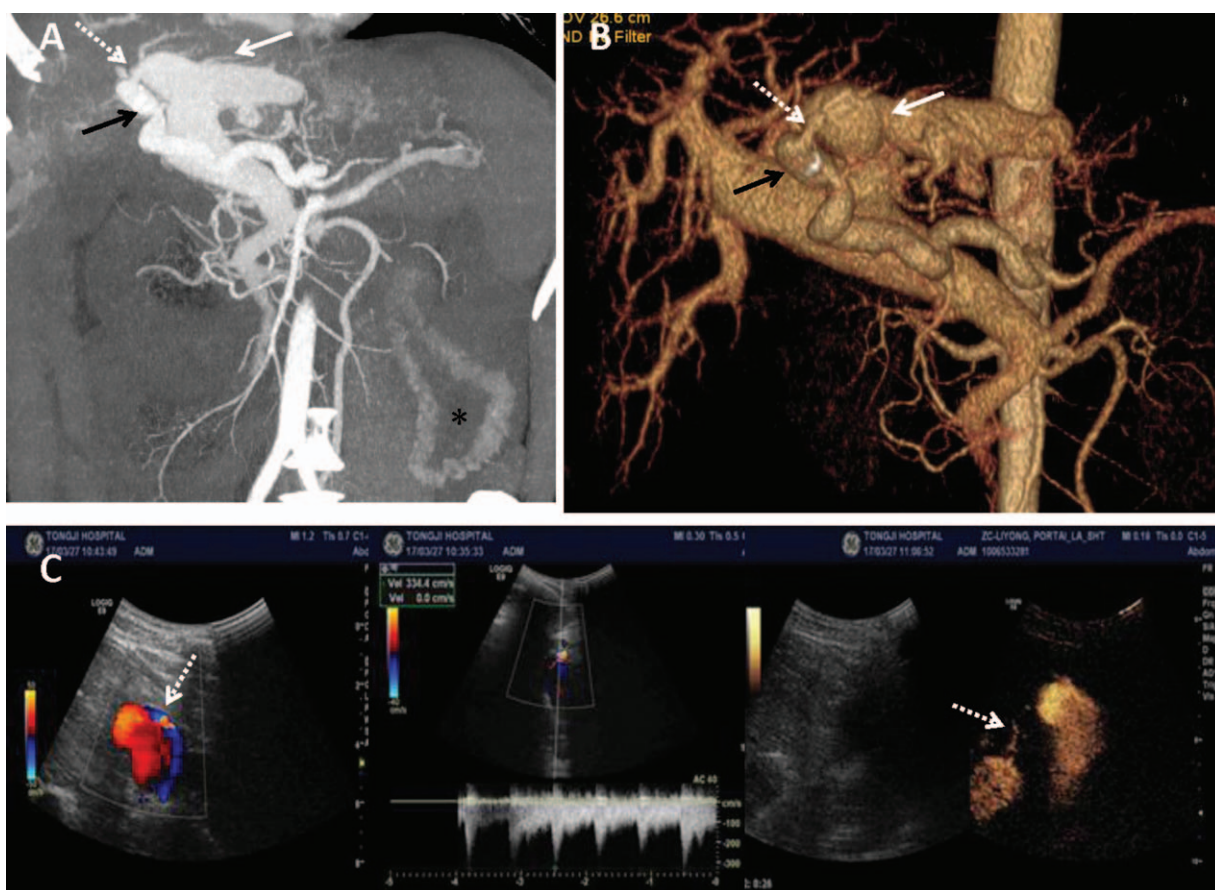


Figure 2. (A and B) Three-dimensional reconstruction CT vividly shows the dilated portal vein (solid white arrows), left hepatic artery (black arrows) and the APF structure (dotted arrows), and diffuse bleeding of the jejunum (black star). (C) Doppler ultrasound shows obvious enlargement of the LPV branch and left hepatic artery. Contrast-enhanced ultrasonography revealed that the portal vein synchronously appeared in the arterial phase, and a high-flow fistula (0.32 cm) from the left hepatic artery near the scar to the LPV branch at a speed of 310 cm/s (dotted arrow). APF = arterioportal fistula, CT = computed tomography, LPV = left portal vein.

in the arterial phase, and a high-flow fistula (0.32 cm) from the left hepatic artery near the scar to the LPV branch was observed at a speed of 310 cm/s (Fig. 2C).

After admission, the patient was treated with a low-protein diet, anti-diarrheic with smectite, antibiotics, and fluid infusion. The stool frequency decreased to about 10 per day. Diagnostic paracentesis was performed. The serum ascite albumin gradient was 24.3 g/L, indicating that the ascites were transudated due to portal hypertension. However, the patient became more and more passive and rejected any injections. On day 6 of hospitalization, the patient's symptoms worsened and he began to vomit persistently with hematemesis, accompanied by abdominal pain, anuria, and shock. His BP level decreased to 75/50 mm Hg.

Laboratory findings showed a hemoglobin level of 79 g/L, white blood cell count of $20.46 \times 10^9/L$, and platelet count of $227 \times 10^9/L$. The kidney function tests showed a blood urea nitrogen level of 60.51 mmol/L, creatinine level of 472 $\mu\text{mol/L}$, and uric acid level of 1286.0 $\mu\text{mol/L}$. Then, the patient underwent hemodialysis treatment 3 times. On day 12 of hospitalization, the patient suffered a large amount of hematemesis that was more than 1000 mL and developed shock. He was treated with blood transfusions and an infusion of terlipressin. Emergency angiography was performed; a 4F diagnostic catheter was inserted into the common hepatic artery via the right femoral artery. A hepatic arteriogram showed a significantly enlarged hepatic proper artery and left hepatic artery, a high-flow connection mainly between the left hepatic artery and the

LPV branch, portalvenous dilatation, and an indistinct right hepatic artery (Fig. 3A). The distal left hepatic artery fistula was embolized with 5 fibered coils (Cook Medical, Bloomington, IN). A postembolization contrast injection showed only a limited residual filling defect of the APF. The left hepatic artery branch and right hepatic artery were clearly displayed (Fig. 3B).

Postoperatively, the systolic murmur over the liver was weakened. However, Doppler ultrasound showed that a high-flow fistula from the left hepatic artery to the LPV branch was still observed at a speed of 284 cm/s (Fig. 4A). After carefully reading the CT images and performing 3-dimensional reconstruction of the hepatic vessels, we found another small fistula between the left hepatic artery and the LPV branch that was less clearly displayed on the CT images (Fig. 4B). This fistula was also embolized with 2 fibered coils and 8 microcoils, followed by a sufficient injection of gelatin sponge particles and 10 mL of lauromacrogol. Control hepatic arteriography at the end of the operation showed complete occlusion of the left hepatic artery and hepatopetal flow in the portal vein (Fig. 5A and B). The systolic murmur over the liver had disappeared. Control Doppler ultrasound showed a thrombosis in the LPV branch; a shunt was not found between the portal vein and left hepatic artery.

During the subsequent days of hospitalization, our patient significantly improved in terms of abdominal distention, abdominal pain, and nausea. A CT scan showed that the ascites and gastrointestinal wall thickening were obviously improved.

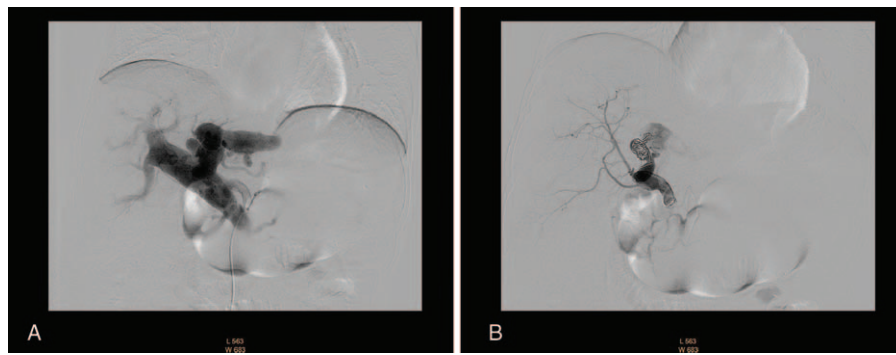


Figure 3. (A) Digital subtraction angiography of the liver and (B) after the first TAE. TAE=transarterial embolization.

The defecation frequency, stool weight, and consistency were nearly normal. The kidney function became almost normal within the next 10 days. The patient was subsequently discharged 2 weeks after the second arterial embolism. During the 6-month follow-up period, the patient remained in remission and was asymptomatic.

3. Discussion

APFs are a rare group of vascular disorders in which the systemic arteries communicate with the portal circulation. Since the first report in 1889 by Goodhart, several causes of APF have been

reported, including congenital vascular malformations.^[9,10] APFs may occur secondary to cirrhosis, hepatic neoplasms, and iatrogenic procedures.^[11] Percutaneous liver biopsy was a main iatrogenic cause of APF, with a 5.4% incidence rate in 93 patients in an original prospective study by Okuda.^[6] A more recent prospective study reported a high APF rate (38%) in 21 patients who underwent hepatic angiography and CT confirmation imaging shortly after biopsy.^[12] The different APF incidence rate between the 2 studies can be attributed to the time interval after liver biopsy and different test procedures.^[13] Noniatrogenic abdominal trauma that is related to APF is rarely reported. Benjamin reported on a 58-year-old man who developed ascites

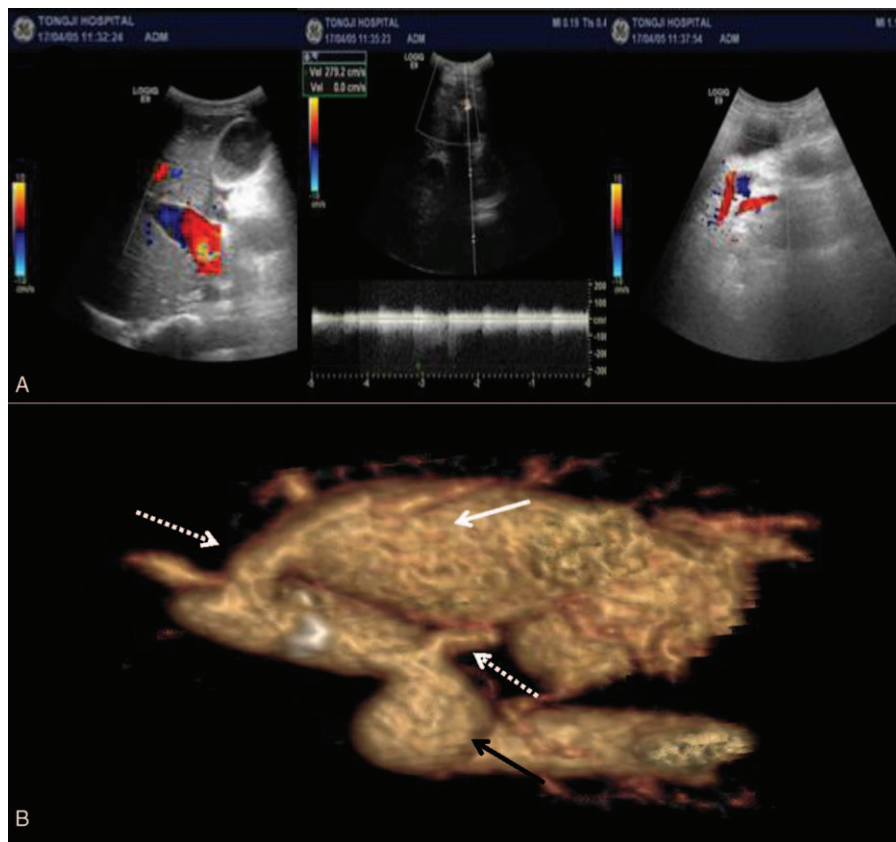


Figure 4. (A) Postoperative Doppler ultrasound still shows a high-flow fistula from the left hepatic artery to the LPV branch. (B) Another small fistula between the left hepatic artery (black arrow) and the left portal vein branch (solid white arrow) is less clearly displayed after the 3-dimensional reconstruction CT images were carefully read (dotted arrow). CT=computed tomography, LPV=left portal vein.

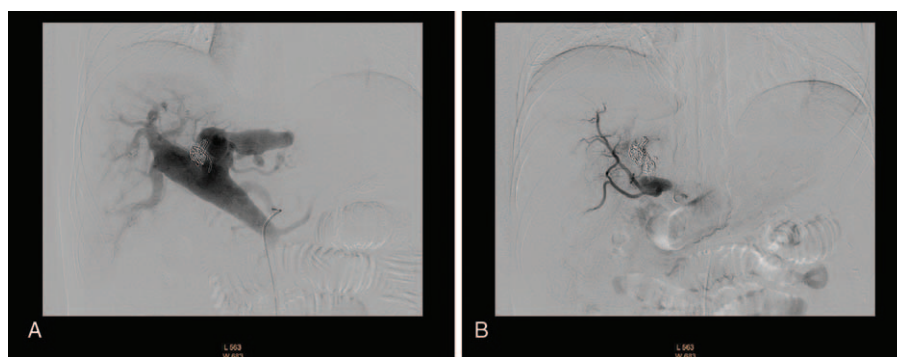


Figure 5. (A) Digital subtraction angiography before and (B) after the second TAE. TAE=transarterial embolization.

that were attributed to an APF 43 years after the liver was lacerated.^[14] In our case, the patient suffered from a penetrating abdominal stab wound 5 years before he developed symptoms.

An APF can be intrahepatic or extrahepatic, depending on the location and volume of shunted blood. Patients with APF may remain asymptomatic or develop severe symptoms, including diarrhea, intestinal ischemia, cirrhosis, or cavernous hemangiomas, or have the symptoms of portal hypertension such as gastrointestinal bleeding or ascites.^[11] A physical examination may reveal the signs of portal hypertension. A continuous murmur over the liver can be heard in about half of patients with this condition. The signs of portal hypertension include continued noninfectious diarrhea, ascites, prerenal oliguria, and anuria, and progressive, severe gastrointestinal bleeding episodes, all of which were the leading signs and symptoms in the present case. Furthermore, the hepatic murmur near the scar facilitated the presumptive diagnosis in our case.

Doppler ultrasound, CT, or digital subtraction angiography (DSA) can be employed to diagnose APF. Doppler ultrasound is typically employed as an initial screening tool, and it can demonstrate an APF if there is frank hepatofugal flow, in which the flow opposite to the direction of flow in the adjacent hepatic artery is seen in the main portal vein or intrahepatic branches.^[15] Limited studies reported on the use of contrast-enhanced ultrasound to detect APF. In our case, contrast-enhanced ultrasound with an intravenous injection of SonoVue not only revealed that the portal vein synchronously appeared in the arterial phase but also clearly revealed a high-flow fistula from the left hepatic artery to the LPV branch. More studies are needed to prove the role of contrast-enhanced ultrasound in diagnosing APF, especially lesions that are not apparent.

A plain CT scan is sufficient to detect ascites and gastrointestinal wall thickening in patients with portal hypertension. However, it is insufficient to show vascular lesions and gastrointestinal hemorrhage. Contrast-enhanced CT can demonstrate enlarged feeding arteries, the location of the fistula, and filling of the portal vein in the early arterial phase. It can also demonstrate associated manifestations, such as esophageal and gastric varices, diffuse bleeding of the gastrointestinal tract, and other arteriovenous malformations.^[16] A recent retrospective study evaluated the performance of multidetector computed tomography (MDCT) in diagnosing APFs in patients with high-grade liver injury.^[17] The MDCT findings of an APF included transient hepatic parenchymal attenuation differences (THPAD); early, increased attenuation of a peripheral or central portal vein compared with the main portal vein; and the “double-barrel” or

“rail tract” signs. In that previous study, THPAD was the most sensitive MDCT finding (82%, 9/11) and had a high specificity (95%, 21/22) to diagnose APF.^[17] Therefore, MDCT should be performed in all patients in whom an APF is suspected.

DSA is the gold standard in the diagnosis, treatment planning, and follow-up of APF.^[18] It is helpful to assess the vascular anatomy to accurately locate the APF and its feeding vessels. It also provides a port of entry for possible embolization.^[19]

Because most fistulae are small, peripheral, or have a low shunt volume, and they can either close spontaneously or remain asymptomatic, the decision regarding whether to treat an APF is divided.^[11] However, with the advent of minimally invasive endovascular therapy, due to the relative safety and effectiveness of this treatment, every APF is considered for treatment, regardless of size or location, to prevent the onset of portal hypertension.

The treatments for an APF are surgical and percutaneous TAE with various materials.^[11] The surgical approaches include surgical ligation of the supplying artery, fistula excision, direct vascular repair, partial hepatectomy, and (rarely) liver transplantation.^[20]

Recently, TAE has been considered the preferable treatment because of its low invasiveness and success in some cases, both in animals and humans.^[21,22] The goal of embolization is not only to perform selective fistula closure but also to preserve the adjacent normal vasculature, which mandates characterization of the collateral sources of the blood supply to the hepatic segments. Materials that are used for embolization include coils, glue, alcohol, and detachable balloons. TAE for an APF is safe and effective overall; however, in some cases, especially in liver transplantation patients, there are multiple feeding arteries, seen in up to 20% to 25% of APF cases.^[23] Smaller APFs were usually ignored or concealed on the initial CT and DSA scans. Embolizing the main feeding artery may expose the secondary feeding vessels, and 3-dimensional reconstruction of the blood vessels is sensitive to show the communication of APFs. Surgeons can perform a less-selective embolization procedure after the initial embolization to ensure that all APFs are shut. In our case, the patient underwent embolization twice because of complicated communication between the portal vein and hepatic artery that was ignored on the initial CT and DSA.

The major risks of TAE are portal venous thrombosis due to reduced flow after embolization, migrated coils, and migrated glue.^[24] These conditions may be prevented by using large devices. Anticoagulant and anti-platelet therapy were discussed in this setting, but no consensus has been achieved yet.^[24]

We described the case of a patient with portal hypertension and gastrointestinal hemorrhage secondary to a traumatic hepatic artery pseudoaneurysm and APF. We successfully treated the patient with coil embolization. This case suggests that taking a detailed medical history, carefully performing a physical examination, and using appropriate imaging methods are necessary to diagnose an APF. Interventional radiology plays an important role in the diagnosis and treatment of an APF; however, as the recurrence rate after coil embolization is unclear, close long-term follow-up is required after treatment.

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