Internal Medicine
The Japanese Society of Internal Medicine
doi: 10.2169/internalmedicine.6565-20
Intern Med 60: 2617-2622, 2021
http://internmed.jp

【 CASE REPORT 】

The First Case of Coil Embolization for Left Gastric Vein Aneurysm with Liver Cirrhosis: A Case Report and Review of the Literature

Seiya Kato[1], Ryotaro Sakamori[1], Ryoko Yamada[1], Kazuhiro Murai[1], Teppei Yoshioka[1], Yuki Tahata[1], Minoru Shigekawa[1], Takahiro Kodama[1], Hayato Hikita[1], Hidenari Hongyo[2], Yusuke Ono[2], Hiroki Higashihara[2], Tomohide Tatsumi[1] and Tetsuo Takehara[1]

Abstract:
We herein report the first case of interventional radiology for a left gastric vein aneurysm with a gastrorenal shunt. The etiology of the aneurysm was considered secondary to portal hypertension and liver cirrhosis due to hepatitis B virus infection. As the aneurysm was asymptomatic but had a tendency to expand, we successfully performed coil embolization for the aneurysm through a gastrorenal shunt.

Key words: portal venous system aneurysms, interventional radiology, coil embolization

(Intern Med 60: 2617-2622, 2021)
(DOI: 10.2169/internalmedicine.6565-20)

Introduction

Portal venous system aneurysms (PVSAs) are considered congenital or secondary to portal hypertension, chronic hepatic disease, trauma, pancreatitis, etc. (1). Among approximately 250 cases of PVSA, only 1 case of a left gastric vein aneurysm was reported (2). Although the need for treatment for PVSAs remains controversial, therapeutic intervention should be considered when aneurysms have symptoms or expansion and show the possibility of rupture, thrombosis or adjacent organ compression (3). Surgical procedures have been the standard treatment for PVSAs, but recently, several cases of successful interventional radiology, such as portal stent graft placement, have been reported.

We herein report the first case of interventional radiology for a left gastric vein aneurysm through a gastrorenal shunt.

Case Report

A 67-year-old woman with hepatitis B virus (HBV)-related liver cirrhosis, hyperammonemia and gastroesophageal varices was undergoing follow-up at our hospital. She had a history of hypophyseal adenoma, hypothyroidism and hyperlipidemia. She was taking entecavir, rifaximin, lactitol, levothyroxine and ezetimibe. She had undergone a follow-up examination every six months with ultrasonography before, and no aneurysm had been detected. Dynamic contrast-enhanced computed tomography (CT) performed for hepatocellular carcinoma surveillance revealed splenomegaly, a gastrorenal shunt, and a saccular left gastric vein aneurysm 22 mm in diameter (Fig. 1A, B). Follow-up CT after 10 months showed enlargement of the aneurysm (32 mm in diameter) with no ascites or portal thrombosis (Fig. 1C, D, 2). Laboratory studies showed anemia (Table 1). The Child-Pugh class was B. The aneurysm was asymptomatic but showed an expanding tendency.

Coil embolization for the saccular aneurysm was performed to prevent rupture. An 8-Fr sheath (ASATO; Medikit, Tokyo, Japan) was inserted via the right femoral vein with local anesthesia. At first, a 4-Fr catheter (Cerulean G; Medikit, Tokyo, Japan) was inserted and placed retrogradely through the tortuous gastrorenal shunt to the left gastric vein with a 0.035-inch guide wire (Radifocus;

[1]Department of Gastroenterology and Hepatology, Osaka University Graduate School of Medicine, Japan and [2]Department of Diagnostic and Interventional Radiology, Osaka University Graduate School of Medicine, Japan
Received: October 26, 2020; Accepted: January 21, 2021; Advance Publication by J-STAGE: March 8, 2021
Correspondence to Dr. Tetsuo Takehara, takehara@gh.med.osaka-u.ac.jp

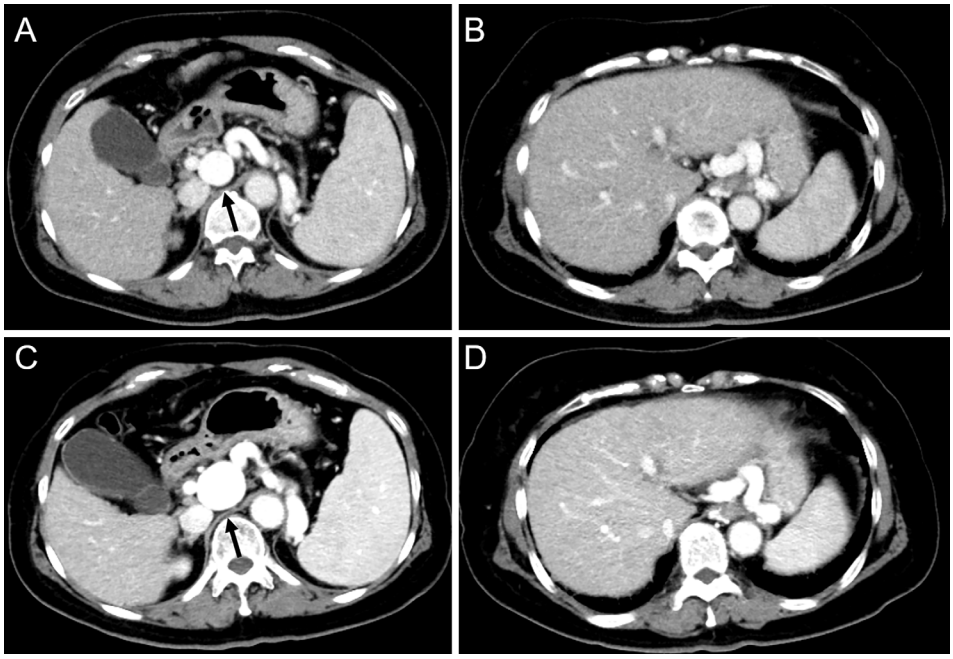

Figure 1. **A, B) Dynamic contrast-enhanced computed tomography (CT) showing splenomegaly, a gastrorenal shunt and a saccular left gastric vein aneurysm (A: arrow). C, D) Follow-up CT after 10 months showing enlargement of the aneurysm (C: arrow).**

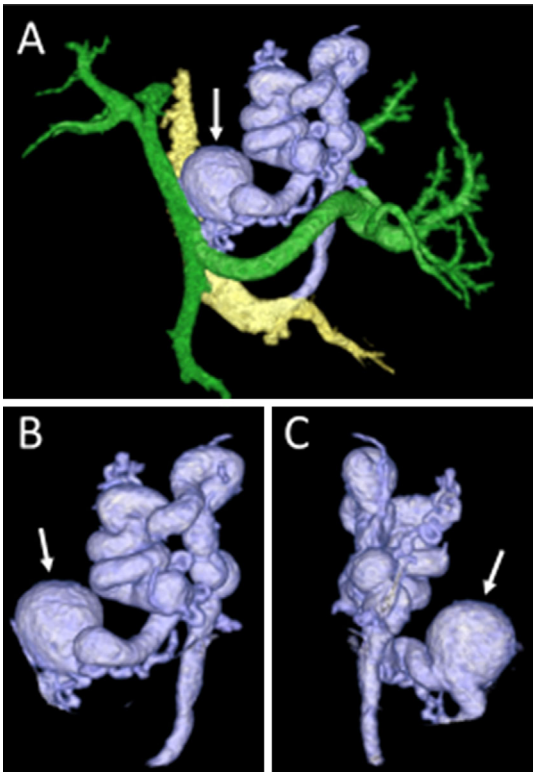

Figure 2. **Multidetector computed tomography images showing the location of a left gastric vein aneurysm. A) Relative location of a left gastric vein aneurysm (arrow) in the portal venous system. Yellow indicates the inferior vena cava and left renal vein. Green indicates the main portal vein, splenic vein and superior mesenteric vein. Purple indicates the left gastric vein aneurysm (arrow) and gastrorenal shunt. B) Ventral side of the left gastric vein aneurysm (arrow) and gastrorenal shunt. C) Dorsal side of the left gastric vein aneurysm (arrow) and gastrorenal shunt.**

Terumo, Tokyo, Japan). A 2.7-Fr coaxial microcatheter (PX SLIM; Penumbra, Alameda, USA) was then advanced into the saccular aneurysm with a 0.025-inch guidewire (Radifocus; Terumo) and a 0.016-inch micro guidewire (Run & Run; Piolax, Yokohama, Japan) (Fig. 3A).

We performed coil packing of the aneurysm, such as via the coil embolization technique, to prevent embolization of the left gastric vein, which was the main collateral vein with portal hypertension. The types and numbers of coils used were as follows: standard Ruby coils, 3 at 36 mm×60 cm, 3 at 28 mm×60 cm, 3 at 24 mm×60 cm, 1 at 14 mm×60 cm and 2 at 12 mm×60 cm; soft Ruby coils, 1 at 10 mm×35 cm and 2 at 8 mm×35 cm; and Penumbra occlusion device (POD) packing coils, 2 at 30 cm and 1 at 15 cm (Penumbra). After coil embolization, venography from the left gastric vein showed the disappearance of blood flow to the aneurysm. Finally, we punctured the right femoral artery and inserted a 4-Fr sheath (Medikit). Arterioportography from the superior mesenteric artery and celiac artery was performed (Fig. 3B, C). We confirmed the disappearance of the blood flow from inside the aneurysm and the preservation of the blood flow to the portal vein and left gastric vein.

Contrast-enhanced CT performed eight days after coil embolization showed neither blood flow to the aneurysm nor portal thrombosis (Fig. 3D-F). The patient was discharged from the hospital 12 days after treatment. After discharge, she had no symptoms, and her liver function did not worsen. The serum NH_3 value at 4 months after treatment was 89 μg/dL, and no symptoms of hepatic encephalopathy were observed. Contrast-enhanced CT at four months after treatment showed neither the blood flow to the aneurysm nor portal thrombosis, and esophagogastroduodenoscopy at four months after treatment did not show any change in the

Table 1. Laboratory Data.

wBC	6,660 /μL	LDH	149 U/L	Alb	3.3 g/dL
RBC	3.42×10^6 /μL	T-Bil	1.4 mg/dL	CRP	0.48 mg/dL
Hb	10.3 g/dL	D-Bil	0.6 mg/dL	NH_3	88 μg/dL
Ht	30.9 %	γ-GTP	33 U/L	Zn	85 μg/dL
Plt	14.5×10^4 /μL	Amy	89 IU/L	AFP	6 ng/mL
PT	62 %	BUN	11 mg/dL	DCP	35 mAU/mL
PT-INR	1.29	Cr	0.64 mg/dL	HBsAg	175.0 U/mL
APTT	32 s	Na	141 mEq/L	HBV DNA	N.D.
D-dimer	1.21 μg/mL	K	3.4 mEq/L	Anti-HCV	N.D.
AST	20 U/L	Cl	105 mEq/L	FIB-4 index	3.38
ALT	8 U/L	TP	7.6 g/dL	ARFI	2.41±0.31 m/s

WBC: white blood cell, RBC: red blood cell, Hb: hemoglobin, Ht: hematocrit, Plt: platelet, PT: prothrombin time, PT-INR: prothrombin time international normalized ratio, APTT: activated partial thromboplastin time, AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactic acid dehydrogenase, T-Bil: total bilirubin, D-Bil: direct bilirubin, γ-GTP: γ-glutamyl transpeptidase, BUN: blood urea nitrogen, Cr: creatinine, TP: total protein, Alb: albumin, CRP: C-reactive protein, AFP: α-fetoprotein, DCP: des-γ-carboxy prothrombin, HBsAg: hepatitis B surface antigen, HBV DNA: hepatitis B virus deoxyribonucleic acid, Anti-HCV: antibodies against hepatitis C virus, N.D.: not detected, FIB-4 index: fibrosis-4 index, ARFI: acoustic radiation force impulse

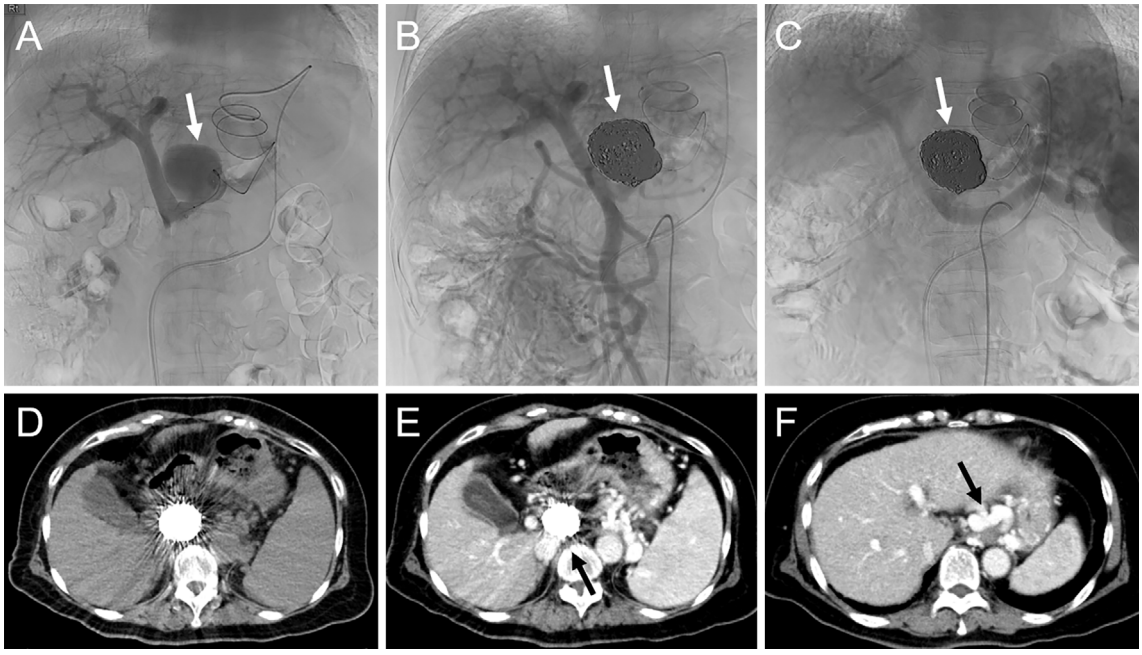

Figure 3. Through a gastrorenal shunt, we performed coil embolization of the left gastric vein aneurysm (arrow). After treatment of the aneurysm, we checked the blood flow of the portal venous system, including the left gastric vein. A) Portography before treatment. B) Arterioportography of the superior mesenteric artery after treatment. C) Arterioportography of the celiac artery after treatment. D) Non-contrast-enhanced computed tomography (CT). E) Dynamic contrast-enhanced CT eight days after treatment, showing neither blood flow in the aneurysm (arrow) nor portal vein thrombosis. F) Remaining blood flow of the gastrorenal shunt (arrow).

gastroesophageal varices (Fig. 4).

Discussion

This is the first case of interventional radiology for a left gastric vein aneurysm through a gastrorenal shunt. In addition, we performed coil embolization for PVSA, which has never been reported, although several cases of portal stent graft placement for PVSAs have been reported.

The portal venous system is a group of veins involved in drainage of the capillary beds of the gastrointestinal tract and spleen into the capillary bed of the liver, including the main portal vein, splenic vein, superior mesenteric vein, inferior mesenteric vein and left gastric vein. PVSAs are rare, and left gastric vein aneurysms are extremely rare. Koc et al. reported that the prevalence of PVSAs was 0.43% among

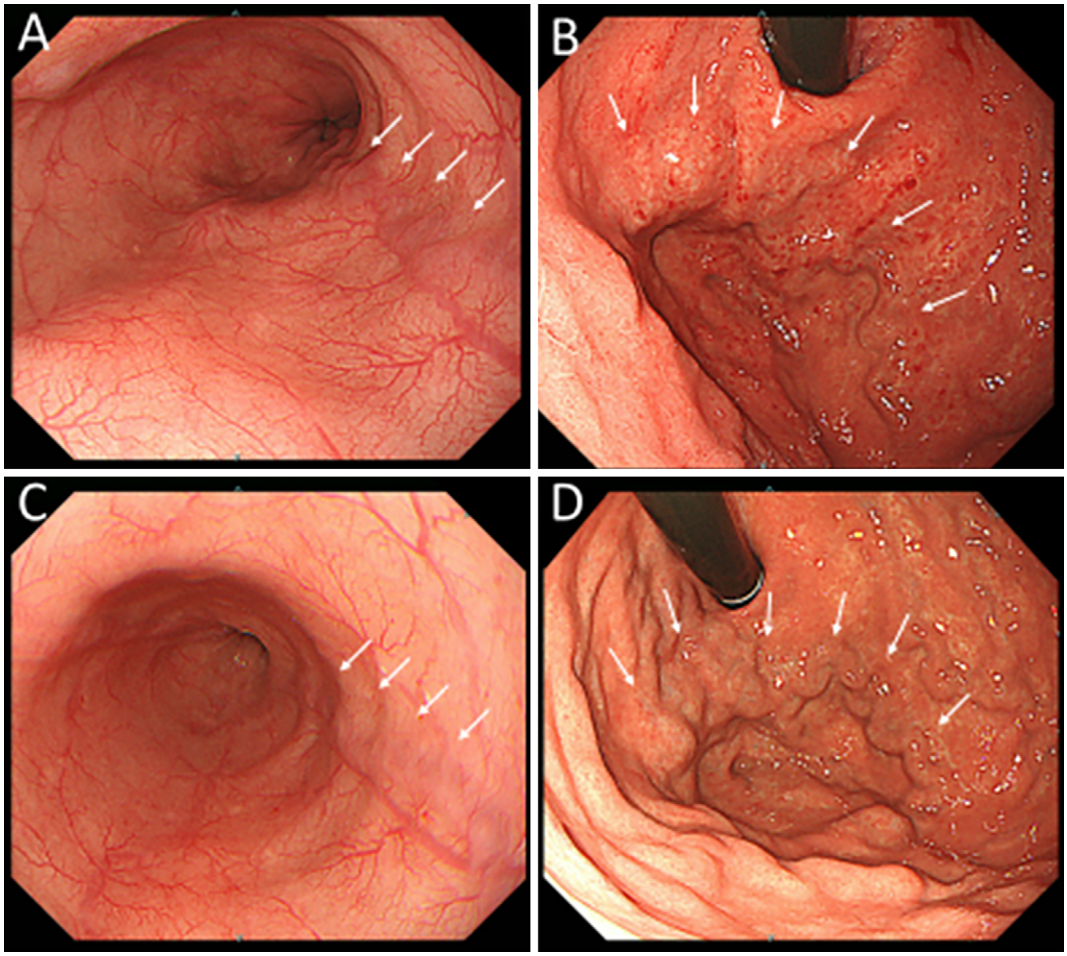

Figure 4. **A, B) Esophagogastroduodenoscopy showing gastroesophageal varices (arrows) before treatment. C, D) Esophagogastroduodenoscopy showing gastroesophageal varices (arrows) four months after treatment.**

4,186 patients who underwent routine abdominal CT (4). The most common locations of PVSAs are the main portal vein and the confluence of the superior mesenteric vein and splenic vein (5). Although approximately 250 cases of PVSA have been reported, only 1 case of a left gastric vein aneurysm has previously reported by Matsutani et al. (2).

The etiology of PVSAs is considered to be congenital or acquired. Congenital causes of PVSAs are considered to include abnormal weakness of the vein wall, incomplete regression of the distal right primitive vitelline vein or branching variants of the portal vein (6, 7). Acquired causes are considered to include portal hypertension, chronic hepatic disease, thrombophilia, trauma, inflammation such as pancreatitis and complications of abdominal surgery (1, 4). Portal hypertension and liver cirrhosis are the main acquired causes, reported in approximately 30% of patients with PVSAs. Recently, the number of patients with PVSAs but without portal hypertension or chronic hepatic disease has increased because of the prevalence of noninvasive medical imaging techniques, such as ultrasound, CT, and magnetic resonance imaging (8, 9). Our patient had liver cirrhosis due to HBV infection, considering the fibrosis-4 (FIB-4) index and acoustic radiation force impulse (ARFI) values (Table 1), and had portal hypertension, considering the existence of splenomegaly, a gastrorenal shunt and gastroesophageal varices. The aneurysm, which was located in a left gastric vein with a gastrorenal shunt, was considered to have been caused by portal hypertension and liver cirrhosis.

The need to treat PVSAs is still controversial. Small aneurysms are usually asymptomatic, while large aneurysms seem to cause symptoms or complications, such as thrombosis, rupture or duodenal or biliary obstruction (10-13). Sfyroeras et al. reported in a systematic review that thrombosis occurred in 13.6%, rupture in 2.2% and adjacent organ compression in 2.2% of 191 cases of PVSA (1). In addition, in 94% of the 53 cases that were followed up, the aneurysm diameter remained stable, and no complications occurred during follow-up. In our case, since the left gastric vein aneurysm was asymptomatic but showed an expanding tendency, we performed treatment, considering the risk of rupture.

Surgical procedures, including aneurysmorrhaphy, aneurysmectomy, splenectomy, splenorenal shunt surgery and distal pancreatectomy, have been the standard treatments for PVSAs (1, 14), and more than 30 cases of surgically treated PVSAs have been reported. However, patients with liver cir-

Table 2. Interventional Radiology for Portal Venous System Aneurysms.

Reference	Age	Gender	Size of Aneurysm (mm)	Location of aneurysm	Etiology	Symptom/ Complication	Treatment
15	56	male	22	splenic vein	trauma	epigastric pain/ rupture	portal stent graft
16	77	female	45	splenic vein	incidental finding after pancreatitis	None	portal stent graft
Our case	67	female	32	left gastric vein	liver cirrhosis	None	coil embolization

rhosis, especially decompensated cirrhosis, such as our patient, have increased rates of perioperative mortality. Recently, several cases of successful interventional radiology for PVSAs, such as portal stent graft placement, have been reported (Table 2), including the treatment of a traumatic splenic vein aneurysm through a transsplenic approach using a portal stent (15) and a splenic vein aneurysm through a transhepatic approach (16). In these two cases, antiplatelet therapy was started after endovascular treatment to prevent portal thrombosis. In our case, the left gastric vein ended at the splenic vein, and the aneurysm was located immediately above the junction of the left gastric vein and the splenic vein. Therefore, we applied coil embolization through a gastrorenal shunt instead of an endovascular stent graft due to the vascular anatomy. We did not start antiplatelet therapy because a stent graft, which can cause clot formation, was not used.

Generally, balloon-occluded retrograde transvenous obliteration (BRTO) is effective for managing solitary gastric varices or recurrent hepatic encephalopathy with a portosystemic shunt (17, 18). Furthermore, there are some data showing that BRTO improves the liver function via an increase in the portal venous flow volume after shunt occlusion (17, 19). Some previous studies have reported that the predictive factor for improvement in the liver function by BRTO was a reduced liver stiffness (20, 21). If we perform BRTO for gastric varices, we need to occlude the gastrorenal shunt, including the left gastric vein aneurysm. However, because the gastric varices form was F1 and her hepatic encephalopathy was controlled by medication, such as rifaximin and lactitol, we decided that BRTO for gastric varices was not necessary at this time. Therefore, we selected coil embolization for the aneurysm. Preservation of the blood flow of the shunt will allow us to perform BRTO through this gastrorenal shunt should the gastric varices grow larger or hepatic encephalopathy become uncontrolled.

In conclusion, to our knowledge, this is the first report of using coil embolization to treat a PVSA. Further studies are needed to evaluate the efficacy of interventional radiology for treating PVSAs.

The patient gave her informed consent for the publication of the details of her case.

The authors state that they have no Conflict of Interest (COI).

References

1. Sfyroeras GS, Antoniou GA, Drakou AA, et al. Visceral venous aneurysms: clinical presentation, natural history and their management: a systematic review. Eur J Vasc Endovasc Surg **38**: 498-505, 2009.
2. Matsutani S, Mizumoto H. Aneurysm of the left gastric vein in a patient with portal hypertension. J Med Ultrason **35**: 141-143, 2008.
3. Calligaro KD, Ahmad S, Dadora R, et al. Venous aneurysms: Surgical indications and review of the literature. Surgery **117**: 1-6, 1995.
4. Koc Z, Oguzkurt L, Ulusan S. Portal venous system aneurysms: imaging, clinical findings, and a possible new etiologic factor. Am J Roentgenol **189**: 1023-1030, 2007.
5. Lopez-Machado E, Mallorquin-Jimenez F, Medina-Benitez A, et al. Aneurysm of the portal venous system; ultrasonography and CT findings. Eur J Radiol **26**: 210-214, 1998.
6. Gallego C, Velasco M, Marcuello P, et al. Congenital and acquired anomalies of the portal venous system. Radiographics **22**: 141-159, 2002.
7. Yang DM, Yoon MH, Kim HS, et al. Portal vein aneurysm of the umbilical portion: imaging features and the relationship with portal vein anomalies. Abdom Imaging **28**: 62-67, 2003.
8. Dognini L, Carreri AL, Moscatelli G. Aneurysm of the portal vein: ultrasound and computed tomography identification. J Clin Ultrasound **19**: 178-182, 1991.
9. Ohnami Y, Ishida H, Konno K, et al. Portal vein aneurysm: report of six cases and review of the literature. Abdom Imaging **22**: 281-286, 1997.
10. Wolff M, Schaefer N, Schmidt J, et al. Thrombosis of a large portal vein aneurysm: treatment by thrombectomy, aneurysmorrhaphy, and portocaval shunt. J Gastrointest Surg **10**: 128-131, 2006.
11. Barzilai R, Kleckner MS. Hemocholecyst following ruptured aneurysm of portal vein; report of a case. AMA Arch Surg **72**: 725-727, 1956.
12. Shimoda M, Kubota K, Sakuma A, et al. Intra-abdominal Hemorrhage Due to Rupture of a Splenic Vein Aneurysm: A Case Report. J Gastrointest Surg **7**: 683-686, 2003.
13. Lall C, Verma S, Gulati R, et al. Portal vein aneurysm presenting with obstructive jaundice. J Clin Imaging Sci **2**: 54, 2012.
14. Cho SW, Marsh JW, Fontes PA, et al. Extrahepatic portal vein aneurysm--report of six patients and review of the literature. J Gastrointest Surg **12**: 145-152, 2008.
15. Kwon OS, Sul YH, Kim JS, et al. Endovascular stent graft for traumatic splenic vein aneurysm via percutaneous transsplenic access. Ann Surg Treat Res **91**: 56-58, 2016.
16. Juscafresa LC, Alfaro MP, Grochowicz L, et al. Endovascular treatment of a splenic vein aneurysm through a transhepatic approach. Diagn Interv Radiol **25**: 166-168, 2019.
17. Kato T, Uematsu T, Nishigaki Y, et al. Therapeutic effect of balloon-occluded retrograde transvenous obliteration on portal-systemic encephalopathy in patients with liver cirrhosis. Intern

Med **40**: 688-691, 2001.

18. Choudhary NS, Baijal SS, Saigal S, et al. Results of portosystemic shunt embolization in selected patients with cirrhosis and recurrent hepatic encephalopathy. J Clin Exp Hepatol **7**: 300-304, 2017.

19. Miyamoto Y, Oho K, Kumamoto M, et al. Balloon-occluded retrograde transvenous obliteration improves liver function in patients with cirrhosis and portal hypertension. J Gastroenterol Hepatol **18**: 934-942, 2003.

20. Yamamoto A, Nishida N, Morikawa H, et al. Prediction for improvement of liver function after balloon-occluded retrograde transvenous obliteration for gastric varices to manage portosystemic shunt syndrome. J Vasc Interv Radiol **27**: 1160-1167, 2016.

21. Ishikawa T, Sasaki R, Nishimura T, et al. Liver stiffness measured by transient elastography as predictor of prognoses following portosystemic shunt occlusion. J Gastroenterol Hepatol **34**: 215-223, 2019.