CASE REPORT

A case of venous aneurysm of a splenorenal shunt

INTRODUCTION
Portosystemic shunts (PSSs) are formed under conditions of portal hypertension due to cirrhosis and frequently associated with hepatic encephalopathy (HE).1 Chronic recurrent HE (CRHE) due to PSS has recently been treated with balloon-occluded retrograde transvenous obliteration (B-RTO).2 In this case, B-RTO was performed for hyperammonemia due to splenorenal shunt with localized aneurysmal change in the splenorenal shunt. Venous aneurysm of the splenorenal shunt (“splenorenal shunt aneurysm”) is rare, and we report herein a case with successful endovascular treatment of a splenorenal shunt aneurysm.

CASE PRESENTATION
Clinical course
A 66-year-old male presented with liver cirrhosis due to non-alcoholic steatohepatitis and hyperammonemia. Follow-up contrast-enhanced CT showed a dilated and tortuous splenorenal shunt and a large venous aneurysm in the shunt. The venous aneurysm showed gradual enlargement over 10 years and worsening hyperammonemia, so balloon-occluded retrograde transvenous obliteration was performed. Under balloon occlusion, 5% ethanolamine oleate was injected from a microcatheter into the venous aneurysm, which was subsequently embolized with microcoils. Contrast-enhanced CT after the procedure showed complete thrombosis of the venous aneurysm. 10 months later, the venous aneurysm reduced in size, and hyperammonemia had improved.

TREATMENT
B-RTO
A coaxial double-balloon catheter system (Candis; Medikit, Tokyo, Japan) was inserted into the splenorenal shunt from the left renal vein via the right femoral vein under local anesthesia. The balloon-occluded retrograde venography showed the portal vein was patent, no thrombosis and the hepatic blood flow was hepatopetal. The microcatheter was advanced into the venous aneurysm (Figure 2A), then 9 ml of 5% ethanolamine oleate (Oldamin; ASKA...
Pharmaceutical, Tokyo, Japan) was injected from the microcatheter under balloon occlusion. Finally, the draining vein was embolized with microcoils (Figure 2B). We used coils of 1.5 times size in diameter compared to the shunt vein for preventing coil migration. A 5-Fr balloon catheter (9 mm diameter, Selecon MP Catheter II; Terumo, Tokyo, Japan) was inserted into the hepatic vein through the right femoral vein, and pressures were measured using a Polygraph MSC-7000 manometer (Fukuda Denshi, Tokyo, Japan). The measured parameters were right atrial pressure, hepatic venous pressure, and wedged hepatic venous pressure (WHVP). WHVP was 22 mmHg. Under balloon occlusion of the splenorenal shunt, WHVP was 32 mmHg. B-RTO was successfully performed, and no complications were observed.

**FOLLOW-UP**

10 months later, the venous aneurysm was seen to have shrunk (Figure 3), and hyperammonemia had improved. No esophageal varices or ascites were noted. Child-Pugh grade changed from B (score 7) to A (score 6) and ALBI grade changed from 2b (score −1.94) to 2a (score −2.47).

**DISCUSSION**

PSSs are common in patients with portal hypertension due to cirrhosis and develop as portal vein pressure increases. These shunts can be divided into intra- and extrahepatic shunts, such as gastrorenal shunt, splenorenal shunt, superior mesenteric vein-inferior vena cava shunt, and inferior mesenteric vein-inferior vena cava shunt, and these can also lead to HE. CRHE is often controlled using drugs such as lactulose or rifaximin, but some cases prove refractory to pharmacotherapy. Surgical ligation is reportedly effective for the treatment of CRHE, but B-RTO has been widely adopted in Japan for the management of HE.

No reports have described cases with localized aneurysmal changes in the splenorenal shunt, but several reports have described cases with HE due to large splenorenal shunt. Venous aneurysms included portal system aneurysm (PSA), and splenorenal shunt aneurysm are very similar in terms of portal hypertension. PSA is associated with not only portal hypertension but also an inherent weakness of the vessel wall. In this case as well, the congenital wall weakness and thinning of the shunt itself were thought to be the main cause of aneurysmal change, with the splenorenal shunt aneurysm subsequently increased by portal hypertension.

Standard treatments for splenorenal shunt aneurysm with HE remain lacking. Careful observation without treatment is often selected for extrahepatic PVA. Surgical treatments for PSA are often indicated in cases with severe symptoms, thrombus formation, worsening of liver function, and enlargement during follow-up. The rupture of PVA has been reported. Sfyroeras et al reported the
diameter of the ruptured PVA was 2 cm.\textsuperscript{9} Similarly, if splenorenal shunt aneurysm continues to increase, there is a risk of rupture. Splenorenal shunt aneurysm should be treated if symptoms such as HE are present or if the aneurysm tends to be large. The increased splenorenal shunt flow is thought to be one of the causes of the aneurysm enlargement and exacerbation of HE. B-RTO is useful to treat the aneurysm itself and HE with splenorenal shunt closure at the same time. In this case, we treated the patient with B-RTO, resulting in thrombosis of the splenorenal aneurysm and shunt closure. The improvement of HE is mainly due to the effect of shunt closure. Thrombosis and reduction of the splenorenal aneurysm by B-RTO will prevent it from rupturing.

Conversely, increased portal blood flow after shunt embolization can lead to complications such as exacerbation of gastric varicose veins, retention of ascites, and progression of hepatic failure.\textsuperscript{10} The indications for treatment of PSS remain unclear, but pre-operative liver function is one of the most important factors in post-operative complications. This case showed Child-Pugh score 7 (class B), and the increase in WHVP was less than 60% before and after balloon occlusion of the splenorenal shunt. No post-operative complications such as varicose vein exacerbation or retention of ascites were observed.

Some recent reports have described portosystemic shunt syndrome, in which the presence of PSS worsens liver function in the long-term.\textsuperscript{11} B-RTO plays a protective role against the lowering of hepatic functional reserve in the long term because portal blood flow increases after B-RTO.\textsuperscript{11,12} In our case, Child-Pugh and ALBI grades changed from Child-Pugh Grade B (score 7) and ALBI Grade 2b (score −1.94) to Child-Pugh grade A (score 6) and ALBI Grade 2a (score −2.47). B-RTO was feasible to improve liver function and to prevent rupture of venous aneurysm.

**CONCLUSIONS**

B-RTO was feasible as a treatment to improve liver function and prevent rupture of splenorenal shunt aneurysm.

**LEARNING POINTS**

- Portosystemic shunt may show aneurysmal formation / aneurysmal change.
- B-RTO for shunt aneurysm was feasible.

**PATIENT CONSENT**

Written informed consent was obtained from the patient for publication of this case report, including accompanying images.

**REFERENCES**

1. Ohnishi K, Nakayama T, Koen H, Saito M, Saito M, Chin N, et al. Interrelationship between type of spontaneous portal systemic shunt and portal vein pressure in patients with liver disease. *Am J Gastroenterol* 1985; 80: 561–4.
2. Watanabe A. Portal-Systemic encephalopathy in non-cirrhotic patients: classification of clinical types, diagnosis and treatment. *J Gastroenterol Hepatol* 2000; 15: 969–79. doi: https://doi.org/10.1046/j.1440-1746.2000.02283.x
3. Philips CA, Rajesh S, Augustine P, Padsalgi G, Ahamed R. Portosystemic shunts and refractory hepatic encephalopathy: patient selection and current options. *Hepat Med* 2019; 11: 23–34. doi: https://doi.org/10.2147/HMER.S169024
4. Waguri N, Hayashi M, Yokoo T, Sato R, Arao Y, Setsu T, et al. Simultaneous combined balloon-occluded retrograde transvenous obliteration and partial splenic embolization for portosystemic shunts. *J Vasc Interv Radiol* 2012; 23: 650–7. doi: https://doi.org/10.1016/j.jvir.2012.01.065
5. López-Machado E, Mallorquí-Jiménez F, Medina-Benítez A, Ruiz-Carazo E, Cubero-García M. Aneurysms of the portal venous system: ultrasonography and CT findings. *Eur J Radiol* 1998; 26: 210–4. doi: https://doi.org/10.1016/S0720-048X(96)01146-1
6. Gallego C, Velasco M, Marcuello P, Tejedor D, De Campo L, Friera A. Congenital and acquired anomalies of the portal venous system. *Radiographics* 2002; 22: 141–59. doi: https://doi.org/10.1148/radiographics.22.1. g02ja08141
7. Koc Z, Oguzkurt L, Ulusan S. Portal venous system aneurysms: imaging, clinical findings, and a possible new etiologic factor. *AJR Am J Roentgenol* 2007; 189: 1023–30. doi: https://doi.org/10.2214/AJR.07.2121
8. Ohnami Y, Ishida H, Konno K, Naganuma H, Hamashima Y, Zeniya A, et al. Portal vein aneurysm: report of six cases and review of the literature. *Abdom Imaging* 1997; 22: 281–6. doi: https://doi.org/10.1007/s002619900190
9. Sfyroeras GS, Antoniou GA, Drakou AA, Karathanos C, Giannoukas AD. Visceral venous aneurysms: clinical presentation, natural history and their management: a systematic review. *Eur J Vasc Endovasc Surg* 2009; 38: 498–505. doi: https://doi.org/10.1016/j.ejvs.2009.03.016
10. Sakurabayashi S, Sezai S, Yamamoto Y, Hirano M, Oka H. Embolization of portal-systemic shunts in cirrhotic patients with chronic recurrent hepatic encephalopathy. *Cardiovasc Intervent Radiol* 1997; 20: 120–4. doi: https://doi.org/10.1007/s002709900118
11. Inoue H, Emori K, Toyonaga A, Oho K, Kumamoto M, Haruta T, et al. Long term results of balloon-occluded retrograde transvenous obliteration for portosystemic shunt encephalopathy in patients with liver cirrhosis and portal hypertension. *Kurume Med J* 2014; 61(1–2): 1–8. doi: https://doi.org/10.2739/kurumenmedj.MS63011
12. Saad WEA, Lippert A, Saad NE, Caldwell S. Ectopic varices: anatomical classification, hemodynamic classification, and hemodynamic-based management. *Tech Vasc Interv Radiol* 2013; 16: 108–25. doi: https://doi.org/10.1053/j.tvir.2013.02.004