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Case Report

Repeated rough coiling technique of portosystemic shunt: A novel treatment for hepatic encephalopathy ☆

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ABSTRACT

Hepatic encephalopathy (HE) usually occurs in the end stages of cirrhosis. During these stages, portosystemic shunt (PSS) is one cause of severe HE. Previous reports have demonstrated that shunt embolization is effective in cases involving a large PSS. However, embolization is risky in some patients because severe ascites and esophageal varices may result from aggravation of portal hypertension. Herein, we report a case in which intentional flow reduction was repeatedly performed for spleno-renal shunt using 2 flow reduction methods, debranching and the rough coiling technique, for a patient with severe HE for whom embolization of the whole PSS pathway was risky. Complete embolization was finally achieved by repeated flow reduction over 5 sessions. The patient tolerated treatment well with no ascites for 4 years after total embolization. If embolization of the whole PSS puts the patient at risk for refractory HE, repeatable flow reduction might provide a good alternative path to single-step embolization.

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Introduction

Hepatic encephalopathy (HE) is a reversible change in consciousness that usually occurs in the end stages of cirrhosis [1]. During these stages, portosystemic shunt (PSS) is a pathway that carries toxins such as ammonia directly into the systemic circulation, and can cause severe HE. A prevalence of 46%-70% for these large portosystemic shunts was identified on routine radiological screening in cirrhotic patients with recurrent HE [2]. PSS also causes enlargement and atrophy of the liver, resulting in a condition called portosystemic shunt syndrome (PSSS) [3,4]. Once HE develops, the prognosis is poor, with a reported 1-year survival rate of 42% and a 3-year survival rate of 23% [5]. HE is generally treated pharmacologically. Many recent reports have found that shunt embolization is effective when the portosystemic shunt is large [2,6–11]. However, while effective, shunt embolization is not always safe to perform for all patients because severe ascites and esophageal varices (EV) can sometimes result from the aggravation of portal hypertension. We report a case in which flow reduction methods were repeatedly performed for SR shunt, including a debranching method and a rough coiling method, for a patient with severe HE who was at high risk from embolization of the entire PSS pathway. Complete embolization was finally achieved by repeated flow reduction methods over 5 sessions.

Case report

A 67-year-old man had been diagnosed with alcoholic cirrhosis 10 years earlier. Rupture of esophageal varices (EV) occurred 5 years prior, resulting in decreased liver function. Ascites was observed after EV ruptured, along with gradually worsening HE. At a previous hospital, he had experienced severe disturbance of consciousness due to severe HE, to the point of becoming bedridden and requiring assistance with eating. His prognosis was poor and he was expected to survive just another 3 months, and was transferred to our hospital for long-term and palliative care because leaving the hospital without intravenous infusion of branched chain amino acids (BCAA) (Aminolevan; Otsuka Pharmaceutical, Tokyo, Japan) was difficult. Blood data on admission showed: total bilirubin (T-bil), 1.0 mg/dL; serum albumin (Alb), 2.8 g/dL; prothrombin time (PT%), 70%; serum ammonia (NH_3), 186 μg/dL; and Child-Pugh score, 9 points. He had already taken diuretics and all available medications for HE. Contrast-enhanced computed tomography (CT) and 3-dimensional (3D) multiphase CT showed cirrhosis, ascites, muscle atrophy, and a large spleno-renal (SR) shunt via a dilated splenic vein (Figs. 1A and B). The SR shunt branched from the splenic vein, divided into 3, merged into one, and joined the left renal vein (Fig. 1C). Doppler ultrasonography (US) showed hepatofugal flow of the portal vein, which was considered to represent end-stage PSSS (Fig. 1D). We considered the SR shunt as one cause of severe HE and decided to embolize the shunt. A 10-Fr sheath was introduced from the femoral vein. We then advanced the 10-Fr double-balloon coaxial catheter consisting of a 10-mm balloon and a 20-mm balloon (Candis system; Medikit, Tokyo, Japan)

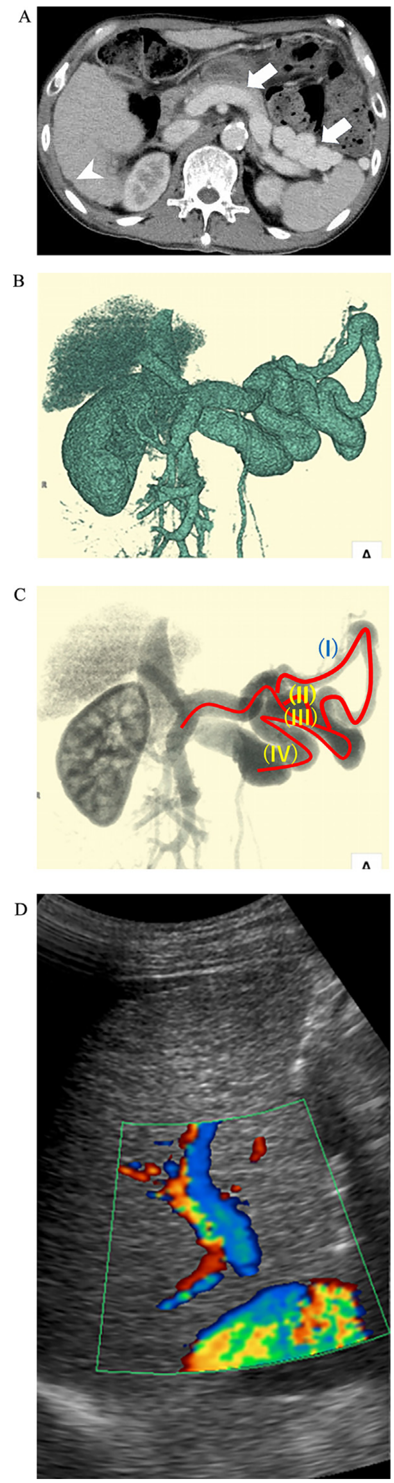

Fig. 1 – (A) Abdominal contrast-enhanced computed tomography (CT) shows hepatofugal flow in a dilated splenic vein (arrow). Slight ascites is observed (arrowhead). (B) Volume-rendered imaging from abdominal dynamic CT shows a large spleno-renal (SR) shunt. (C) Schematic images of the SR shunt. The SR shunt branched from the splenic vein, divided into 3 (I-III), merged into one (IV), and joined the left renal vein. (D) Doppler ultrasonography shows hepatofugal flow in the portal vein (red signal).

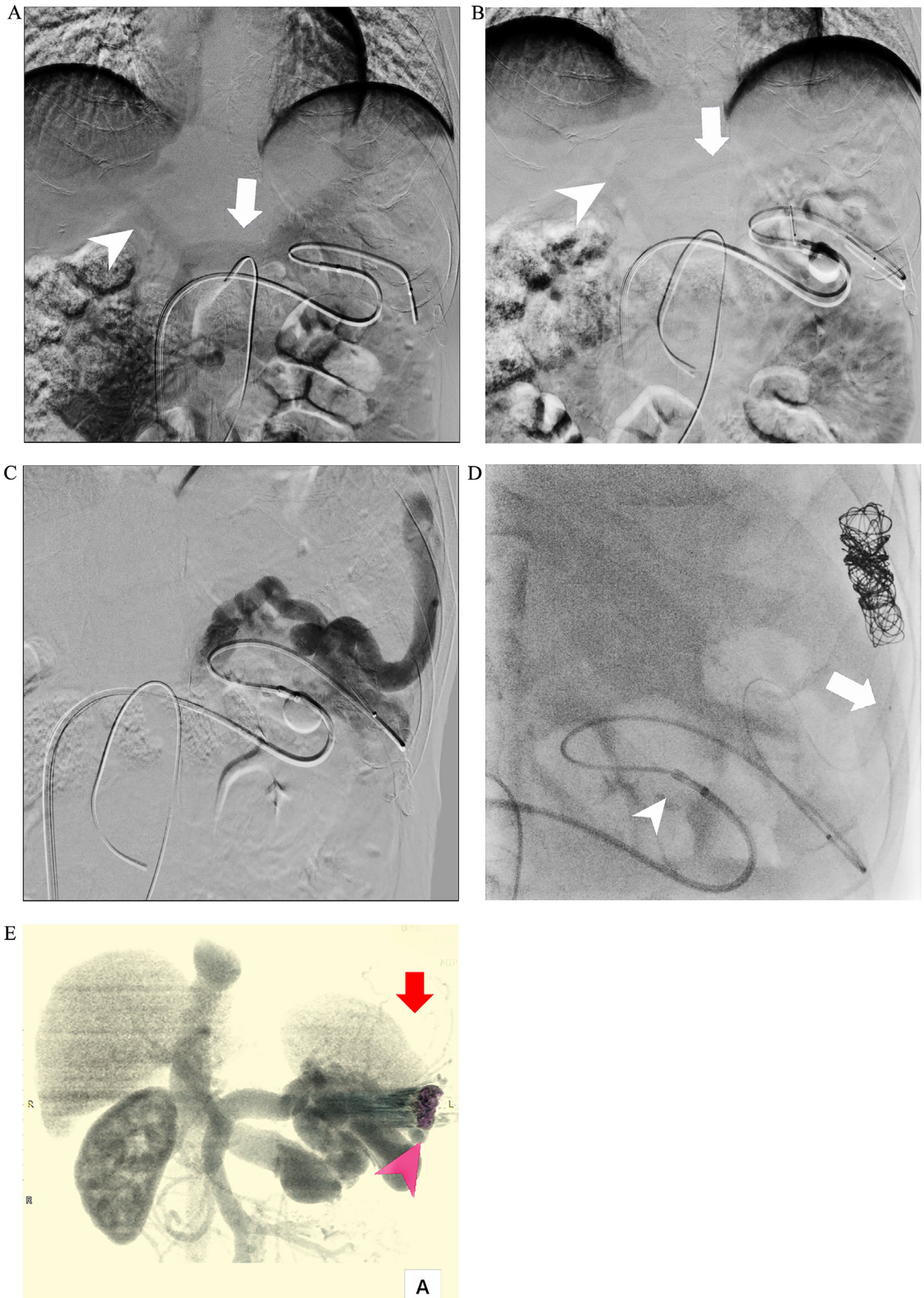

Fig. 2 – (A) Portography via the superior mesenteric artery (SMA) shows hepatofugal flow in the splenic vein (arrow) and portal vein (arrowhead). (B) SMA portography under balloon occlusion of the splenorenal (SR) shunt. (C) Balloon-occluded retrograde transvenous venography (BRTV) from the SR shunt. The 3 divided shunts shown in Fig. 1C are visualized. (D) Coil embolization of one of 3 shunts shown as (I) in Fig. 1C Microcatheter was inserted (arrow) and 2-cm balloon was positioned in the SR shunt (arrowhead). (E) Volume-rendered images from abdominal dynamic CT after coil embolization. One of the 3 shunts (shown as (I) in Fig. 1C) has disappeared (arrow). Seven inserted coils are visualized (arrowhead).

into the SR shunt via a left renal vein. We added a 5-Fr, 10-mm balloon catheter (Terumo, Tokyo, Japan) and measured wedge pressure of the right hepatic vein to confirm portal pressure. In addition, we inserted the 4-Fr catheter into the superior mesenteric artery (SMA) to perform intra-arterial portography. SMA portography showed a dilated splenic vein and large SR shunt (Fig. 2A). We then temporarily occluded the SR shunt with a Candis balloon catheter and performed a balloon occlusion test (measurement of shunt pressure and portography). SMA portography did not show any vessels such as the portal or splenic vein due to congestion (Fig. 2B). Wedge hepatic vein pressure (WHVP) elevated from 15.4 to 37.5 mmHg (143% elevation) under balloon occlusion test in the SR shunt. We considered the patient's situation with current intake of oral diuretics, presence of ascites, failure to visualize the portal vein in the shunt occlusion test, wedge hepatic vein pressure exceeding 30 mmHg with an increase of over 50% (143%) under the shunt occlusion test, and embolization of the SR shunt as a potential risk for refractory ascites after shunt embolization. We therefore avoided total occlusion of the SR shunt in a single procedure. We adopted a treatment strategy of gradually reducing blood flow in the SR shunt over multiple sessions. This step-by-step embolization was considered a repeatable method of flow reduction. After balloon-occluded retrograde transvenous venography (BRTV), we then inserted a microcatheter system into one of the 3 separate SR shunts and performed coil embolization using 7 metallic coils (Azur CX18; Terumo). This technique is named the debranching technique (Figs. 2C-E). After the procedure, serum NH_3 had decreased to 119 μg/dL. Because this procedure was well tolerated and symptoms of HE improved slightly, we performed the second embolization using the debranching technique after 1 month to embolize one of the 3 collaterals with 5 micro-coils (Fig. 1C; vessel II). One month after the second embolization, a third embolization was performed. After the last 2 embolizations, 3 SR shunts became one on BRTV (Fig. 3A). Shunt pressure remained high, elevating from 22 to 35 mmHg (59% elevation) under balloon occlusion testing. We considered that total embolization of the SR shunt was still risky and performed flow reduction of the SR shunt again. We carefully placed 4 coils (Ruby coils; Penumbra, Alameda, CA) using a rough coiling technique to avoid complete shunt embolization while reducing blood flow in the SR shunt. After coil installation, blood flow was slowed but not stopped (Fig. 3B). After the procedure, serum NH_3 was slightly decreased and symptoms gradually improved. With a total of 3 procedures during 3 months of hospitalization, the patient regained consciousness, was able to eat unaided, and was able to walk and start rehabilitation. Finally, the patient, whose prognosis had been estimated as 3 months, was discharged home without intravenous BCAA infusion 5 months after the first procedure. During outpatient follow-up, the scheduled 4th and 5th flow reduction procedures were performed over 10 months with 7 coils using a rough coiling technique in the same manner as the third procedure. His condition stabilized, but after outpatient blood testing showed hyperammonemia (260 μg/dL), he was re-admitted to the hospital for a final complete embolization. We re-inserted the balloon catheter into the SR shunt to perform BRTV (Fig. 4A). We inserted 10 more coils and 5 mL of 5% ethanolamine oleate with iopamidol (EOI) to embolize the SR shunt. We confirmed complete embolization (Fig. 4B). Figure 4C showed a total of 36 coils after these procedures. Finally, hepatopetal portal flow was visualized after total embolization (Fig. 4D). After embolization, serum NH_3 had decreased to within normal range. Doppler US showed hepatopetal flow, where hepatofugal flow had been observed before starting treatment (Fig. 5A). The patient remains well without ascites, although minor hemorrhage from EV was observed 2 years after complete embolization. Ammonia level had gradually decreased after the first procedure (Fig. 6). Body weight was 60 kg with a 10-kg gain compared to before treatment because of improvement of nutritional

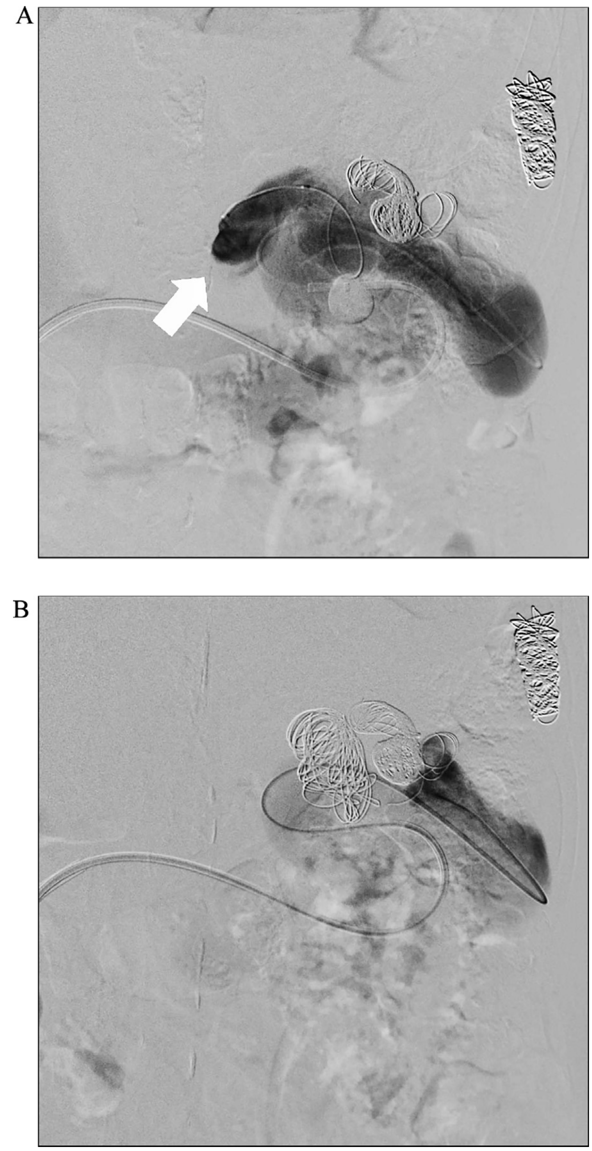

Fig. 3 – (A) Balloon-occluded retrograde transvenous venography (BRTV) from the SR shunt. The splenic vein close to the portal vein is observed in a retrograde manner (arrow). The SR shunt now consists of one vessel after embolization of 2 of the 3 initial vessels (Fig. 1C, I and II). (B) Flow reduction after insertion of 4 coils. The SR shunt is not embolized.

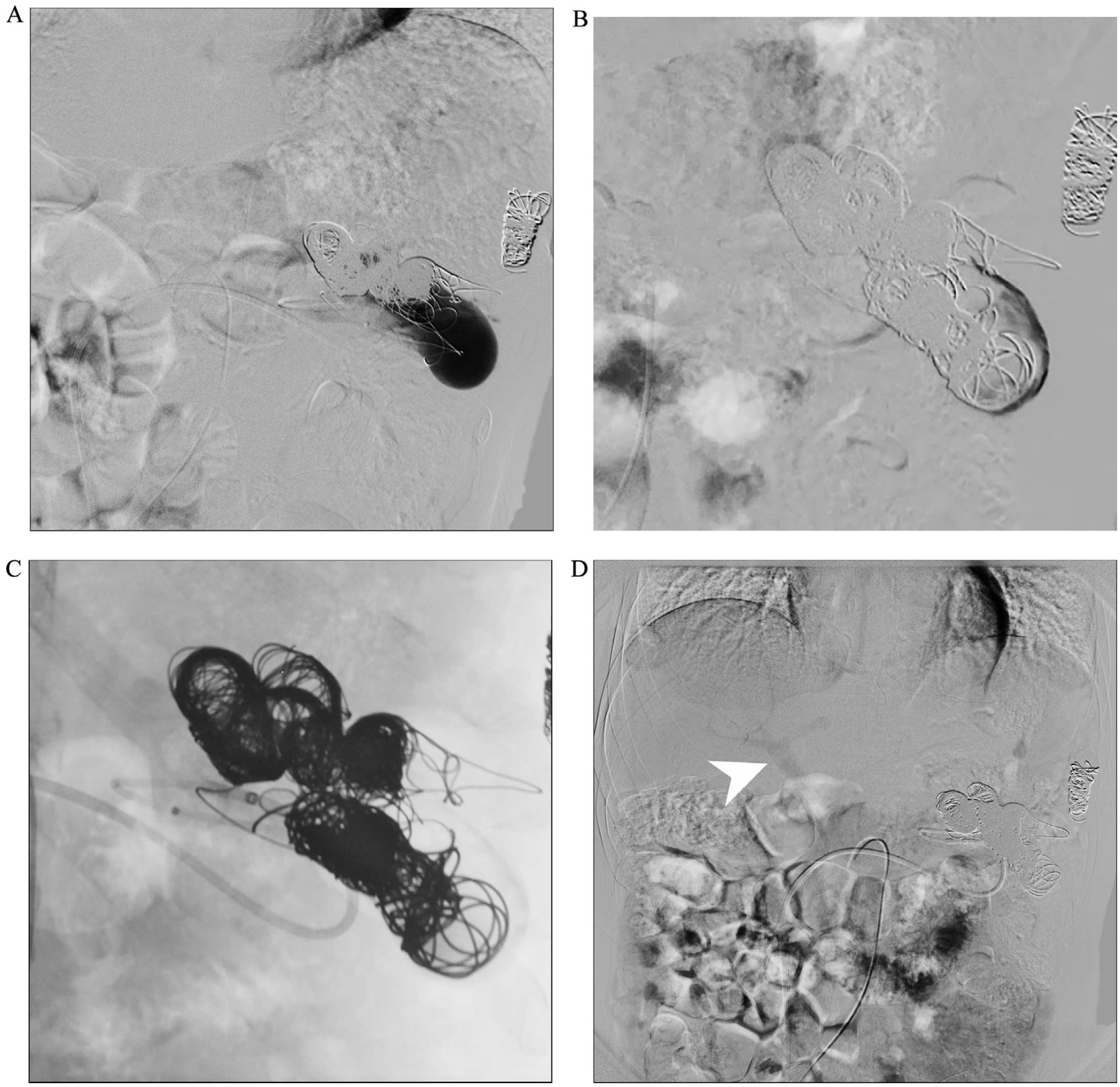

Fig. 4 – (A) Balloon-occluded retrograde transvenous venography (BRTV) from the patent SR shunt. (B) SR shunt after total embolization. Stasis of contrast media is observed near the coils. (C) Thirty-six coils were inserted in 5 procedures. (D) Portography via the superior mesenteric artery (SMA). Hepatopetal flow of the portal vein is observed (arrowhead). The splenic vein is not seen because of total embolization of the SR shunt.

status and muscle thickening through movement. CT 4 years after the first procedure showed no ascites and thickened muscles due to his improved abilities to eat and move (Fig. 5B).

Discussion

The effectiveness of complete embolization of a large PSS has been widely reported [10]. Shunt embolization to treat refractory HE has proven effective and safe in selected patients [10]. Fifty-nine percent of patients treated by embolization remain symptom-free by 3 months later [7]. In 37 patients, 48.6% remained HE-free over a follow-up period of approximately 700 days. A recent randomized trial showed the effectiveness of treatment with findings such as increased liver volume, increased serum albumin, and reduced levels of serum ammonia after embolization [2]. In this case, the patient showed gradual improvements in HE during multiple shunt flow reductions using methods including debranching and the rough coiling technique. Finally, total shunt embolization was performed without complication. HE improved, disappearing after total shut embolization.

The clinical presentation of this patient revealed 2 important clinical implications. First, we applied 2 kinds of flow reduction technique. The debranching technique (Fig. 7A) can be used in patients with multiple shunts, embolizing one and leaving the other patent. In this case, we applied debranching for the first and second procedures. The rough coiling technique (Fig. 7B) is used for patients with a single vessel for

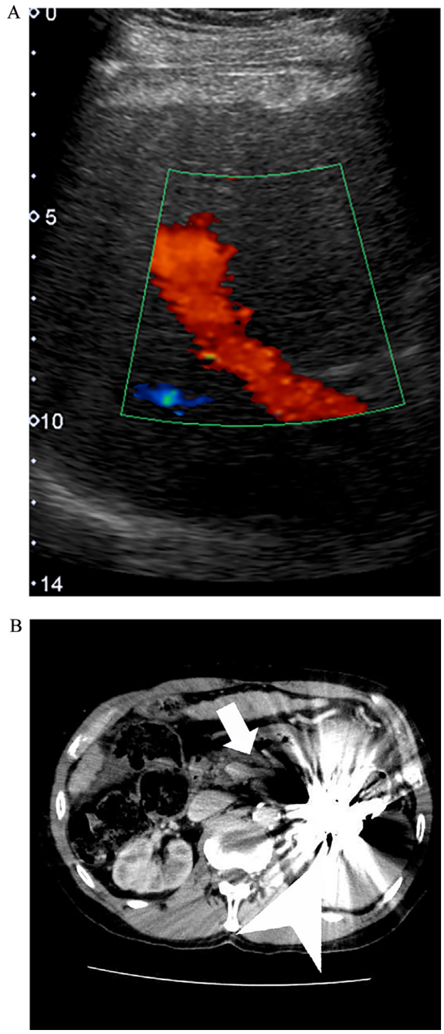

Fig. 5 – (A) Doppler ultrasonography shows hepatopetal flow in the portal vein (red signals). (B) Abdominal contrast-enhanced computed tomography (CT) of the abdomen. The splenic vein is narrowed compared to preoperative CT (Fig. 1A; arrow). Coil artifact in the splenorenal (SR) shunt is observed (arrowhead).

the PSS and involves rough insertion of a few coils to avoid completely embolizing the shunt. We applied this method in the third to fifth procedures. To the best of our knowledge, this is the first report of this kind of repeated flow reduction technique for embolization of a PSS In particular, the rough coiling technique has not been described in previous studies. Although symptoms in our patient only showed mild improvement following each procedure the procedures were well tolerated and could be performed safely and repeatedly. Krinisch et al. reported a different method for partial closure using a diabolo-shaped covered stent created using 2 different stents [12]. However, that technique needs insertion of the long-covered stent, which is sometimes difficult to insert into a small or tortuous PSS. Our method can be performed via microcatheter, which can be applied to a wide variety of PSSs.

Second, although embolization of a PSS is effective, complications due to worsened portal hypertension after embolization remain problematic. No studies have reported results for large numbers of patients regarding contraindications for embolization of the PSS in PSSS patients with HE. As for effectiveness, Laleman et al. identified the Model for End-Stage Liver Disease (MELD) score as the strongest predictor of HE recurrence, with a cutoff of 11 for patient selection [7]. As for safety, Philips et al. reported a Child-Pugh score >11 as predictive of mortality in patients with HE [8]. One of the severe complications is ascites, including refractory ascites and worsening of EV [2,8]. Obviously, patients with refractory ascites or large EV are not considered as candidates for embolization. When WHVP on occlusion testing of the PSS by balloon catheter is ≥30 mmHg, we currently perform a sensitive determination of the indications for shunt occlusion, as reported by Kanazawa et al. in shunt ligation for congenital portal vein closure [13]. A WHVP after shunt closure of <22.8 mmHg has been reported as safe [6,14]. Regarding the rate of increase in portal vein pressure, surgical shunt closure should reportedly be indicated when liver function is good and the rate of increase due to trial closure of the shunt should be limited to 50%-55% [15,16]. Knirsch et al. reported that if WHVP during balloon occlusion testing increases above 15 mmHg, they do not perform embolization for children [12]. In this case, although the Child-Pugh score was 9, WHVP was 35 mmHg (>30 mmHg),

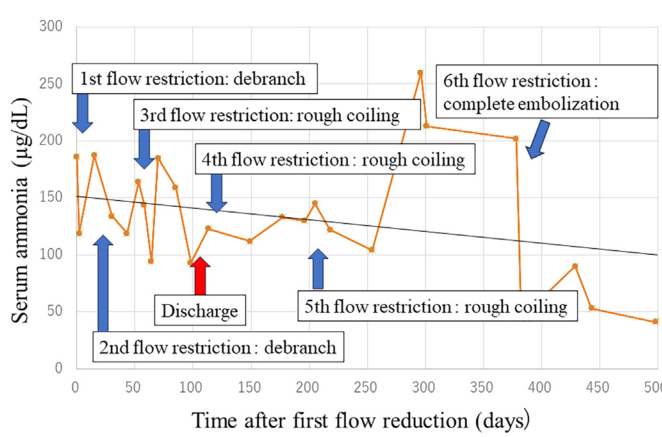

Fig. 6 – Changes in serum ammonia (µg/dL) over time from the first procedure. Serum ammonia gradually decreases over the 5 procedures.

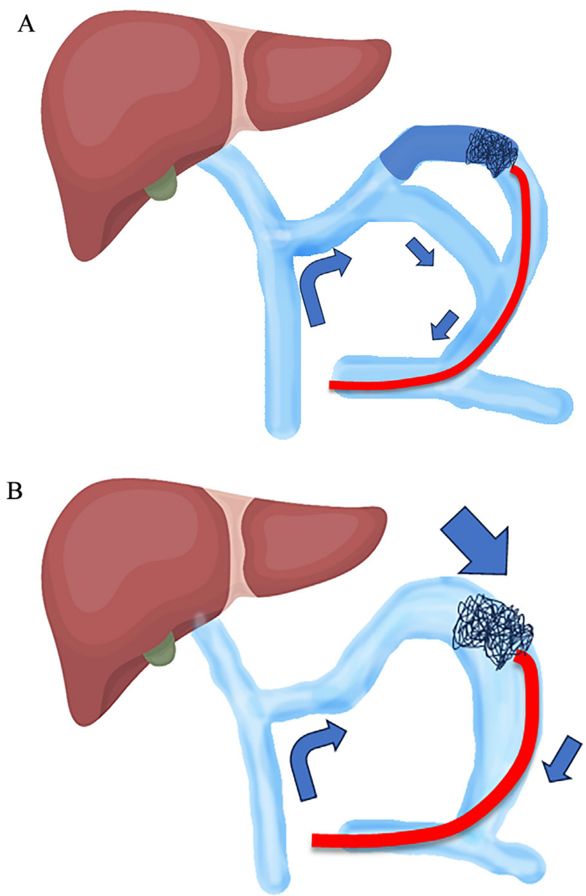

Fig. 7 – Schematic explanations of the 2 methods. (A) Debranching technique: If 2 or more shunts are present, one is completely occluded, leaving the others. (B) Rough coil technique: If one shunt is present, blood flow is reduced by inserting coils roughly into a single shunt, taking care not to completely embolize the vessel.

and the value and rate of increase was 22.1 mmHg and 143% (15 mmHg and >50%, respectively). Total shunt embolization was considered risky. In the case of surgical shunt closure, a 2-step closure is recommended with partial occlusion to initially acclimatize the intrahepatic portal system to increased flow, with complete occlusion performed 6-12 months later [17]. The flow reduction method in this case by catheter was based on a similar concept to this 2-step closure. After repeated flow reduction, the large shunt could be embolized safely and the patient achieved a good clinical course without HE or ascites. This approach may be one option for patients for whom single-step closure may be too risky.

Conclusion

In this report, we described successful embolization of the PSS using repeated flow reduction methods with a debranching technique and a rough coiling technique. Symptoms of HE resolved after embolization without emergent side effects or refractory ascites. If patients are at risk with total embolization of the PSS for refractory HE, this repeated flow reduction method might be an option for embolization.

Patient consent

Regarding this case report, written, informed consent for publication was obtained from the patient.

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