Abstract
Stomal varices represent a rare and serious complication among patients with portal hypertension and external enteric diversion. These varices often cause severe anemia due to recurrent bleeding. We successfully treated stomal varices using a transcatheter intrahepatic portosystemic shunt (TIPS) and endovascular repair for post-procedural hepatic encephalopathy with a stent-in-stent technique.

Key words: stomal varices, transcatheter intrahepatic portosystemic shunt (TIPS), stent-in-stent technique

Introduction
Stomal variceal hemorrhage is a rare and serious complication among patients with both portal hypertension and external enteric diversion. Some authors have reported that transcatheter intrahepatic portosystemic shunt (TIPS) is a less invasive method with a favorable rebleeding rate as compared with other treatment modalities. However, TIPS often induces hepatic encephalopathy.

We report a case in which recurrent stomal variceal hemorrhage was successfully treated with TIPS placement and post-procedural hepatic encephalopathy was resolved using a stent-in-stent technique.

Case Report
A 65-year-old woman with bleeding from a sigmoid colostomy was admitted to our hospital. On admission, vital signs were unstable. Hemoglobin level was 11.3g/dL and platelet count was 49,000/μL. Blood biochemistry showed: albumin, 3.1g/dL; aspartate aminotransferase, 65IU/L; alanine aminotransferase, 31IU/L; total bilirubin, 1.1mg/dL; and prothrombin activity, 70%. The patient had a medical history of chronic hepatitis C, Child-Pugh class B, and abdominoperineal resection for recurrent rectal cancer, after which she had experienced recurrent melena. Contrast-enhanced abdominal computed tomography (CT) demonstrated stomal varices (Fig. 1a), massive ascites (Fig. 1b), and enteral congestion due to portal hypertension (Fig. 1c).

Transhepatic portography showed the stomal varices passing through the inferior mesenteric vein (Fig. 1d). Stomal variceal bleeding due to liver cirrhosis with portal hypertension was diagnosed. Anemia did not improve despite repeated blood transfusions. Portosystemic shunt was therefore considered necessary to stop recurrent stomal variceal bleeding. We considered that surgical treatments such as portosystemic shunt surgery or complete revision of the stoma would likely exacerbate liver cirrhosis, so TIPS was selected as the less invasive method for creating the portosystemic shunt.

TIPS was performed as follows. The portal vein was punctured with a Rosch-Uchida transcatheter liver access kit (Cook, Bloomington, Indiana, United States) (Fig. 2a). The TIPS tract was created between the right hepatic vein and intrahepatic portal...
vein (Fig. 2b) and dilated with a balloon catheter. An 8 × 40-mm metallic stent (Luminexx stent; C. R. Bard, Murray Hill, New Jersey, United States) was placed in the tract (Fig. 2c). After the procedure, portal vein pressure decreased from 30 to 27 mmHg. Six days after the procedure, CT revealed disappearance of stomal varices (Fig. 3a), massive ascites (Fig. 3b), and enteral congestion (Fig. 3c). The patient was discharged without any complications 8 days after the procedure.

Six weeks after the procedure, she was admitted to our hospital with hepatic encephalopathy. Her serum ammonia level was 273 μg/dL, compared to 85 μg/dL the day after the TIPS procedure. She was administered branched chain amino acids (BCAAs), but symptoms remained poorly controlled. Improvement of hepatic encephalopathy required reduction of shunt flow through the TIPS route, so we reduced the inner lumen of the TIPS route. We inserted a sheath introducer through the right jugular vein to approach the TIPS route, then deployed a 10 × 40-mm stentgraft (Fluency; C. R. Bard, Murray Hill, New Jersey, United States) within the previously inserted stent (stent-in-stent technique). After this procedure, the ammonia level decreased to 120 μg/dL without BCAA administration, and hepatic encephalopathy resolved. The patient experienced no further recurrence of bleeding episodes until her death 2 years after the stent-in-stent technique due to progression of rectal cancer.

Discussion

Bleeding from stomal varices is a rare and serious complication of portal hypertension of patients with chronic liver failure and a permanent stoma. The varices arise at the border of the mucocutaneous junction of the stoma as a result of spontaneous anastomoses between the high-pressure portal circulation and the relatively low-pressure systemic venous system. Since the original description by Resnick et al.3 in 1968, numerous treatment strategies have been reported. Conte et al.4 reviewed 71 cases of stomal hemorrhage and reported the various treatment methods
and respective efficacy rates. They concluded that surgical shunt placement represents the most effective means of treatment, but also noted that “porto-caval shunting should be restricted to these patients who can tolerate major surgery and who have a reasonable life expectancy”. Such surgical procedures may have a lower incidence of both rebleeding and need for additional procedures, but are associated with high post-operative morbidity and mortality rates. Cello et al.5 found that elective surgical mortality rates in patients with Child-Pugh class C cirrhosis were as high as 50%. Liver transplantation is a definite treatment option for recurrent stomal variceal bleeding with decompensated liver cirrhosis, but in Japan, patients with cancer are not accepted as recipients for liver transplantation.

TIPS is considered a less invasive method for the creation of a intrahepatic portosystemic shunt, and is also effective against bleeding stomal varices. Given the problems associated with surgical shunts
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and liver transplantation, we consider TIPS as the preferred treatment for stomal variceal bleeding.

However, major concerns remain regarding the use of TIPS in the treatment of stomal variceal bleeding, namely: 1) long-term patency of the TIPS tract; and 2) the risk of hepatic encephalopathy. Reocclusion rates with TIPS may vary depending on the situation and reocclusion could be managed by repeat TIPS or balloon dilatation of the occluded stent lumen. The duration of TIPS patency has been reported as 5 to 48 months in the literature. In our case, the stent lumen remained patent for 2 years. In the presence of decompensated liver function, TIPS may sometimes result in secondary hepatic encephalopathy or liver failure, as seen in our case. In general, the symptoms of hepatic encephalopathy are mild and improve with administration of BCAAs. However, we could not control the hepatic encephalopathy with administration of BCAAs. We then inserted a stent-graft into the previously inserted stent (stent-in-stent technique), and succeeded in controlling hepatic encephalopathy. Fanelli, et al, reported inserting a covered stent-graft into the previously inserted stent increased friction and turbulence caused by the interposed stent meshes, and reduced shunt blood flow. As a result, hepatic encephalopathy was resolved. We decided the size of inserting stent based on this study and succeeded in resolving hepatic encephalopathy. Hemorrhage of stomal varices is most effectively managed by reducing the underlying portal hypertension. TIPS is a less invasive, less morbid means of portal decompression than open portocaval shunting and appears to offer similar effectiveness. The rebleeding rate after TIPS compares very favorably with other treatment modalities. We recommend TIPS as the preferred method for treating stomal varices.

**Conflict of Interest disclosure**
The authors declared no conflicts of interest.

**References**