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Transjugular intrahepatic portosystemic shunt for repeated bleeding of hemorrhoids caused by severe portal hypertension with ectopic varices: A case report

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ABSTRACT

Background: Severe portal hypertension is life-threatening and can bring adverse complications such as ascites, gastroesophageal varices, and edema. It can, even cause variceal hemorrhage, which may lead to a high risk of death. There is a rare incidence in bleeding of hemorrhoids caused by severe ectopic varices.

Case presentation: We report the case of a female patient with a 20-year history of hepatitis B virus infection who presented with repeated bleeding of hemorrhoids caused by severe portal hypertension with ectopic varices that is connection between the superior mesenteric vein and rectal venous plexus. Laboratory results revealed a hemoglobin level of 74 g/L. Finally, the patient was successfully treated with transjugular intrahepatic portosystemic shunt (TIPSS) placement without variceal embolization after a multidisciplinary comprehensive opinion. In the two-month follow-up period, the patient had failed to develop hepatic encephalopathy or hematochezia, and computed tomography venography (CTV) indicated that the stent was unobstructed and ascites disappeared.

Conclusions: TIPSS placement is effective for the case, and we hope this case can help improve clinicians' awareness of hemorrhoidal bleeding with severe portal hypertension. Portal hypertension should also be considered during the diagnosis and treatment, as opposed to hemorrhoidal bleeding alone. Moreover, abdominal CTV is recommended as an effective imaging examination method to determine the stent status after operation.

Background

Portal hypertension (PH) is the main cause of mortality and complications in liver cirrhosis.¹ It can lead to the formation of collateral circulation between the portal vein and systemic circulation, like esophagogastric varices (EGV), abdominal wall varices, and spontaneous splenorenal shunt.² Ectopic varices caused by severe PH account for only 5% of all variceal bleeding, and the most common locations are surgical stomas, duodenum, jejunum-ileum, and colon.³ However, it is extremely rare that the diagnosis of PH with ectopic varices is confirmed due to hemorrhoidal bleeding. Furthermore, the treatment of ectopic variceal rebleeding compared with that of their initial bleeding is crucial because of the high mortality.⁴ According to the guidance,⁵ endoscopic variceal ligation (EVL) is the preferred treatment for variceal bleeding caused by PH. However, there is no consensus on the treatment of ectopic varices,

which requires a complete knowledge of the vascular anatomy and multidisciplinary approach. Therefore, we report the case of a female patient with repeated bleeding of the hemorrhoids caused by severe PH with ectopic varices. She was successfully treated with a transjugular intrahepatic portosystemic shunt (TIPSS). We hope this case can help improve clinicians' awareness of hemorrhoidal bleeding with severe portal hypertension, and realize the efficacy of TIPSS placement, which can alleviate the clinical symptoms and reduce the occurrence of bleeding significantly.

Case presentation

On January 1, 2019, a 71-year-old female patient was referred to our hospital due to severe hematochezia (bright red blood) for 2 days. She had no other clinical manifestations, such as dizziness, headache, and

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dyspnea. Her medical history mainly included nearly two decades of hepatitis B virus (HBV) and mixed hemorrhoids. The patient was not previously treated with EVL. Conservative treatment improved her symptoms during this hospitalization.

On February 2, 2019, the patient was readmitted to our hospital because of rebleeding of hemorrhoids for 1 day. She had slight dizziness, five-day diarrhea, and no other clinical manifestations other than perianal hemorrhoid prolapse. Subsequent enhanced computed tomography (CT; SOMATOM Sensation 64 Spiral, Siemens, Munich, Germany) showed a nodular filling defect, approximately 1.7 cm in diameter, on the main portal vein trunk in the portal venous phase and significant dilation on the hemorrhoid venous plexus in the venous phase. In addition, splenomegaly, a large amount of ascites, and PH were noted (Fig. 1). Her laboratory results revealed: HBV infection; red blood cell (RBC) and platelet counts $2.03 \times 10^{12}/L$ and $66 \times 10^9/L$, respectively; hemoglobin, albumin, and total bilirubin levels 74 g/L, 3.6 g/dL, and 0.77 mg/dL, respectively; prothrombin time (PT), 18.2s; and international normalized ratio, 1.53. Serum tumor-related markers or alpha-fetoprotein levels were generally unremarkable. The patient was rated as B according to the Child–Turcotte–Pugh score.

During this hospitalization, the patient was provided with expanded blood volume and electrolyte balance therapy. Because of severe PH, there was still an increased risk of rebleeding. Therefore, TIPSS placement was finally considered after the comprehensive opinions of gastroenterologists, interventional radiologists and anorectal specialists.

The procedure was performed under heparinization and local anesthesia. First, the right internal jugular vein was punctured using the Seldinger technique. A 10-F catheter sheath was introduced into the hepatic vein, and the free hepatic vein pressure (FHVP) was measured at 2.84 mmHg. Then, we successfully punctured the right portal vein trunk using RUPS-100 (Cook Incorporated, Indiana, USA). After confirmation with direct portal vein angiography, a 5-F pigtail catheter (Cook Incorporated, Indiana, USA) was introduced into the main portal vein, and the portal pressure was measured at 27.21 mmHg. To assess the hepatic perfusion pressure, the portal pressure gradient (PPG) was easily calculated at 24.37 mmHg.⁶ Severe ectopic varices of the superior mesenteric vein–rectal venous plexus and mild EGV were clearly seen (Fig. 2). The punctured channel between the hepatic and portal veins, including the liver parenchyma and vessels, was expanded with an 8 cm \times 6 mm balloon (Bard Peripheral Vascular, Inc., Arizona, USA). Subsequently, an 8 cm \times 8 mm bare metal stent (Bard Peripheral Vascular, Inc., Arizona, USA) and 5 cm \times 8 mm coated stent (W.L. Gore & Associates, Inc., Arizona, USA) were placed in this channel. Finally, reexamination revealed that the portal venous blood flowed smoothly back into the inferior vena cava through the stent (Fig. 3). The portal venous pressure and FHVP were remeasured at 19.12 and 9.02 mmHg, respectively, and the PPG was 10.10 mmHg. Compared with the previous one, the PPG was significantly decreased, so the embolization of varices was not performed.

Postoperatively, the patient's condition was closely observed. Albumin supplementation and electrolyte balance therapy were still performed. In addition, low-molecular-weight heparin sodium was used for anticoagulant therapy. Symptoms such as dizziness, diarrhea and hematochezia obviously improved; meanwhile, the laboratory results such as the RBC, hemoglobin level and PT were slightly better than before. Ascites and lower limb edema gradually reduced. The patient made a smooth recovery and was discharged in good condition 4 days after the operation. Nearly 2 months later, computed tomography venography (CTV) revealed that the implanted stent remained in its original proper position without a blocking or migration, and ascites, variceal hemorrhage and EGV were not seen (Fig. 4). Neither hepatic encephalopathy (HE) nor recurrence of hematochezia was reported in the follow-up period.

Discussion

It is reported that rectal varices frequently occur in PH with cirrhosis, but only few patients become symptomatic.⁷ Hemorrhoids are prevalent in our population; according to statistics, the vast majority of people have asymptomatic hemorrhoids, and patients with frequent hematochezia are relatively rare.⁸ The varicose rectal venous plexus plays a significant role in the formation of hemorrhoids, the minority of which may be formed due to cirrhosis with PH. If the portal pressure is significantly high, the risk of hemorrhoidal bleeding may be increased. Hence, if patients experience repeated bleeding of hemorrhoids in the recent period, it is necessary for them to be examined and receive treatment timely.

Henry et al⁹ have reported that the formation of ectopic varices is associated with the congenital vascular anatomy and PH. Patients with congenital vascular abnormalities may have ectopic varices to compensate with the presence of PH. In the case of a patient with PH, the portal vein blood backflow had been blocked for a long time, and blood stasis had been maintained in the rectal venous plexus because of the connection between the superior mesenteric vein and rectal venous plexus. When the portal pressure significantly increased, it led to the hemorrhage of rectal venous plexus, that is, hemorrhoid hemorrhage. After bleeding for a certain period of time, the portal pressure would slightly decrease due to the low blood volume, and hemorrhoid hemorrhage may be relieved. Rebleeding easily occurred when the portal pressure rose again. Accordingly, it is essential to improve the quality of life and control bleeding for senile patients. TIPSS placement was finally performed due to the multidisciplinary proposal.

At present, TIPSS placement is the second-line treatment for gastrointestinal bleeding due to cirrhosis with PH, and mainly used for patients with a high risk of rebleeding and the circumstance that EVL is invalid. The Baveno VI consensus¹⁰ and practice guidelines⁵ suggest that for patients at high risk of rebleeding or with active bleeding, early TIPSS can significantly control bleeding and reduce the incidence of rebleeding. Additionally, TIPSS is better than other treatment in improving ascites

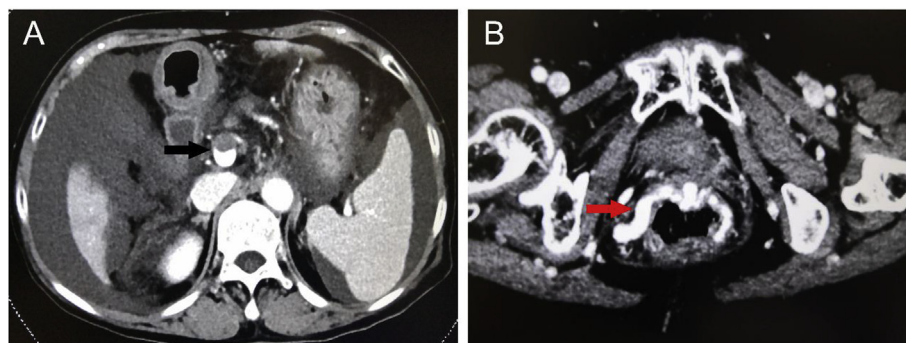


Fig. 1. A. Contrast-enhanced CT showed a nodular filling defect on the main portal vein trunk (black arrow), splenomegaly and a large amount of ascites. B. Hemorrhoid varicosity (red arrow).

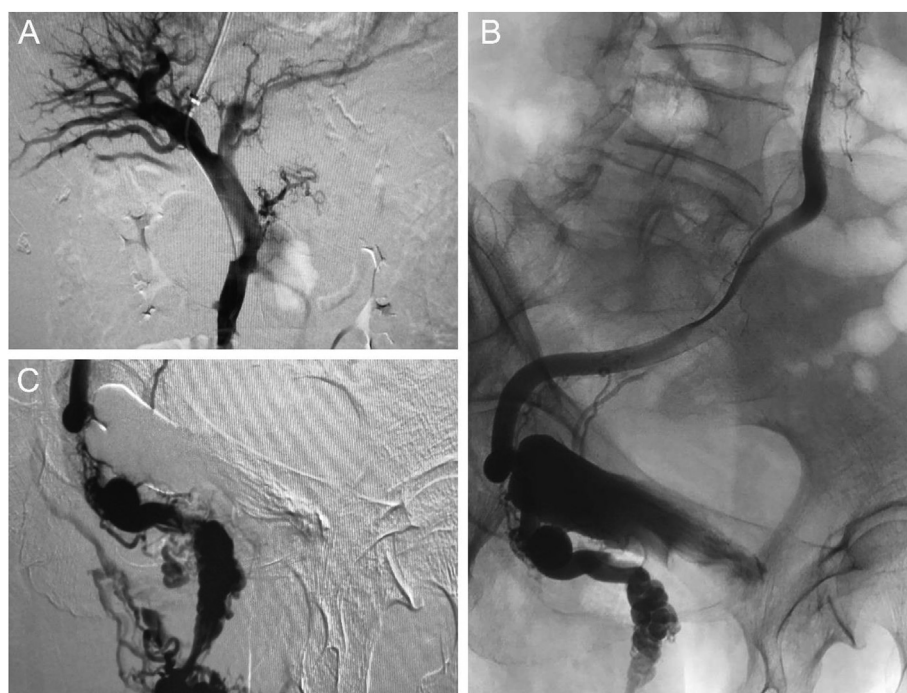


Fig. 2. A. Direct portal vein angiography. B. Severe ectopic varices of superior mesenteric vein–rectal venous plexus. C. Hemorrhoid varicosity.

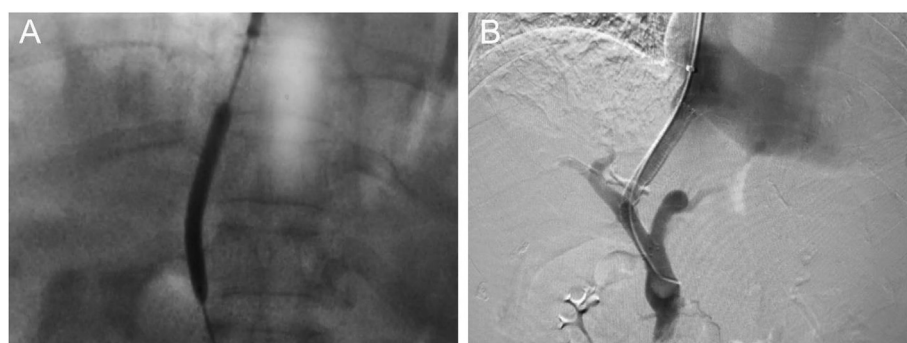


Fig. 3. A. The punctured channel expanded with an 8 cm × 6 mm balloon. B. The portal blood flowed smoothly back into the inferior vena cava through stent.

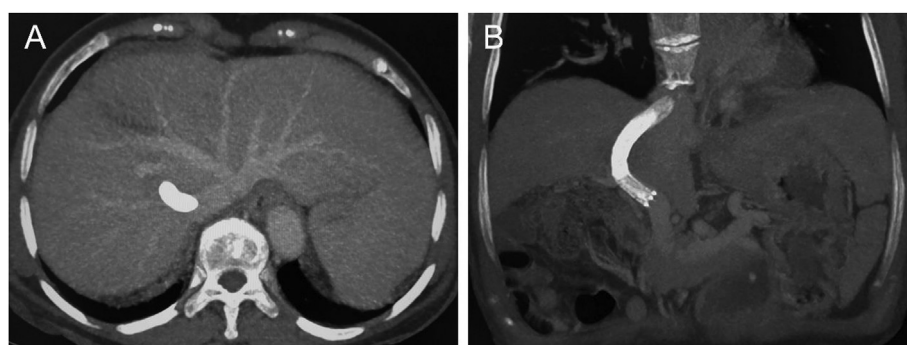


Fig. 4. Two months after the operation, computed tomography venography showed that ascites disappeared and stent remained in its original proper position without blocking.

and relieving. Postoperative complications, such as edema, HE¹¹ and melena, can be treated with medication and diet therapy.

Nonetheless, ectopic variceal hemorrhage is life-threatening and its treatment is complicated and crucial. We recommend TIPSS placement for repeated bleeding of ectopic varices, and the treatment of combining

with embolization should be considered according to the diameter of varices, amount of bleeding and postoperative PPG. We hope this case can help improve clinicians' awareness of hemorrhoidal bleeding with severe PH. In addition, abdominal CTV is recommended as an effective imaging examination method to determine the stent status after the

operation.

Declaration of competing interest

The authors declare that they have no conflicts of interests to this work.

Abbreviations

PH	portal hypertension
EGV	esophagogastric varices
EVL	endoscopic variceal ligation
TIPSS	transjugular intrahepatic portosystemic shunt
HBV	hepatitis B virus
CT	computed tomography
RBC	red blood cell
PT	prothrombin time
FHVP	free hepatic vein pressure
PPG	portal pressure gradient
CTV	computed tomography venography
HE	hepatic encephalopathy

Ethics approval and consent

A case report is intended to develop information to be shared for medical and educational purposes. Both institutional review board approval and patient consent for this case were obtained.

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