

# Transjugular intrahepatic portosystemic stent shunt placement and embolization for hemorrhage associated with rupture of anorectal varices

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## Abstract

Portal hypertension can lead to ectopic varices, which occur most frequently in the rectum. Rectal variceal bleeding in patients with portal hypertension is rare but can be life-threatening if not diagnosed and treated in a timely manner. However, no specific treatment guidelines have been established for rectal variceal bleeding. We herein report a case involving a woman with portal hypertension due to autoimmune liver disease who was successfully treated with a transjugular intrahepatic portosystemic stent shunt and variceal embolization. We recommend treatment of refractory ectopic variceal bleeding with a transjugular intrahepatic portosystemic stent shunt in combination with embolization.

## Keywords

Hemorrhoid venous bleeding, ectopic varices, embolization, transjugular intravenous portosystemic shunt, portal hypertension, cirrhosis

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## Introduction

Portal hypertension can result in the formation of collateral circulation between the portal vein and systemic circulation.<sup>1</sup> Although portal hypertension commonly causes esophageal variceal bleeding, it can also cause varicose vein bleeding at other

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sites (i.e., ectopic varicose vein bleeding).<sup>1</sup> Bleeding of rectal varices in patients with portal hypertension is rare, with an incidence of 1% to 8%.<sup>2,3</sup> The most recent survey in Japan showed that ectopic varicose veins are most common in the rectum, accounting for 44.5% of all ectopic varicose veins.<sup>4</sup> However, patients with portal hypertension rarely develop rectum varicose vein bleeding. Confirmation of the diagnosis is difficult, and the bleeding is sometimes severe. It can be life-threatening if it is not clearly diagnosed and correctly treated in timely manner. Additionally, no treatment guidelines have been established for rectal variceal bleeding. We herein report a case of hemorrhoid varicose vein bleeding caused by portal hypertension that was successfully treated by timely diagnosis and application of a transjugular intrahepatic portosystemic shunt (TIPS) and embolization. This case is being reported to remind clinicians that portal hypertension should be considered in patients with hematochezia and that TIPS in combination with embolization is necessary for refractory ectopic variceal bleeding.

## Case report

Both patient consent and institutional review board approval for this case report were verbally obtained.

In April 2010, a 60-year-old Chinese woman diagnosed with autoimmune liver disease and decompensated cirrhosis of the liver was admitted to Peking Union Medical College Hospital because of abdominal distention. Her medical history included hemorrhoids. Conservative medical treatment improved her hematochezia. The patient had esophageal varices, which were treated twice with endoscopic variceal ligation.

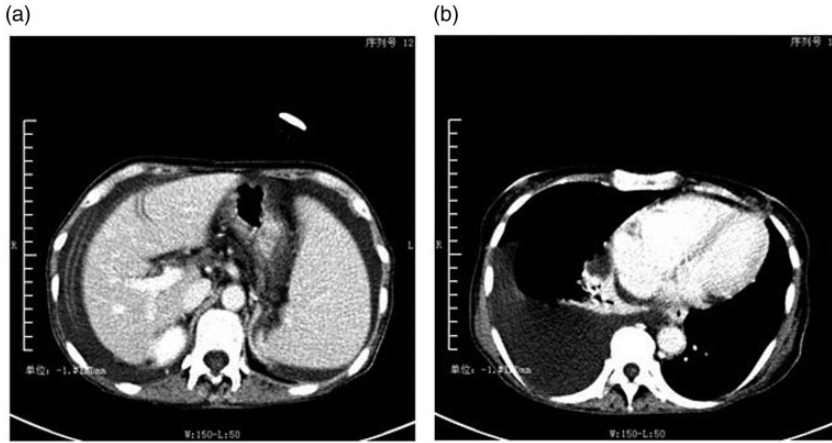
On 22 December 2014, the patient was admitted to our hospital because of a 1-week history of diarrhea. A physical examination revealed that the patient's spleen was

located 4 cm under her ribs. Laboratory tests showed autoimmune hepatitis-related antibodies, a red blood cell count of  $3.02 \times 10^{12}/L$ , a hemoglobin level of 90 g/L, and a platelet count of  $17 \times 10^9/L$ .

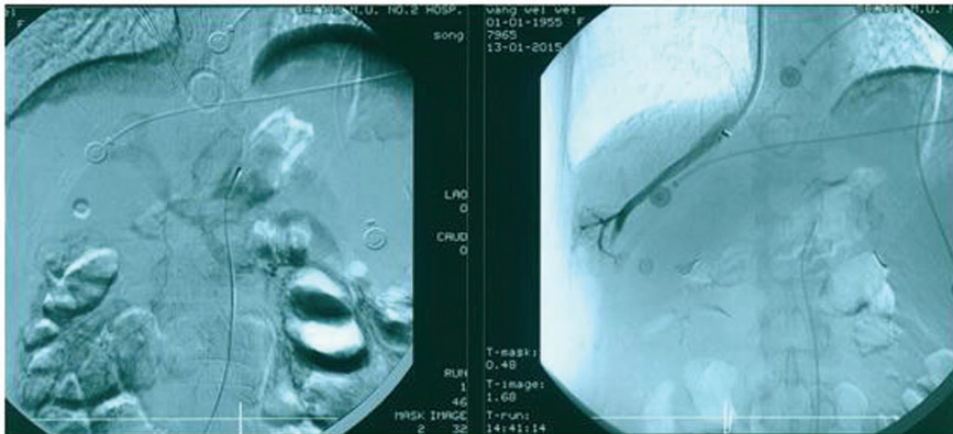
The patient's Child-Pugh class was C. No obviously abnormal tumor-related antigen or alpha-fetoprotein levels were found. A computed tomography scan showed cirrhosis of the liver, splenomegaly, ascites, and portal hypertension (Figure 1). The patient was provided parenteral nutrition. She developed a high fever and asthma symptoms, so the possibility of respiratory failure was considered. The patient was transferred to the intensive care unit where she received mechanical ventilation, anti-inflammatory therapy, pleural effusion drainage, and an aminosalicic acid enema.

On 4 January 2015, the patient developed hematochezia. Blood clots were found in the lumen of the rectum, and lower gastrointestinal hemorrhage was suspected. Medication to reduce gastric acid and somatostatin to reduce the portal pressure were administered, and the patient received a transfusion of plasma, red blood cells, and platelets. The hematochezia ceased after this treatment. On 13 January 2015, the patient passed stool containing fresh blood and underwent the same above-mentioned treatment. During treatment, her heart rate increased to 130 beats/min and her blood pressure decreased to 60/40 mmHg. Because of the patient's cirrhosis, portal hypertension was thought to be the cause of the rectal varices. Therefore, we considered TIPS placement in combination with variceal embolization as an appropriate intervention for this patient.

On 13 January 2015, a TIPS was embedded under local anesthesia using a RUPS-100 puncture set (Cook Medical, Bloomington, IN, USA) and indirect portal vein angiography (Figure 2). A 6-F arterial sheath and 5-F vertebral artery



**Figure 1.** Computed tomography and multiple-slice computed tomographic portography showing portal hypertension and hydrothorax.

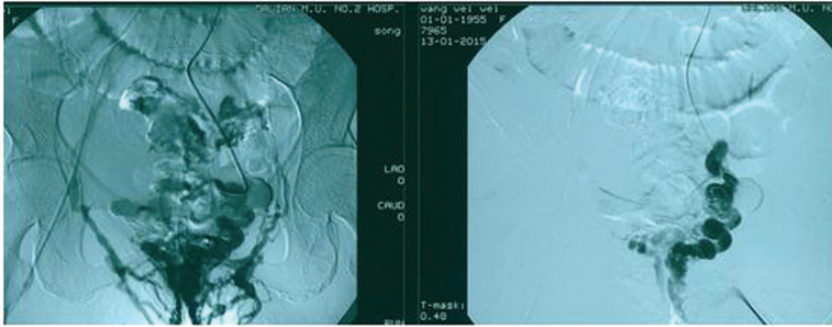


**Figure 2.** Indirect portal vein angiography with the RUPS-100 system (Cook Medical, Bloomington, IN, USA).

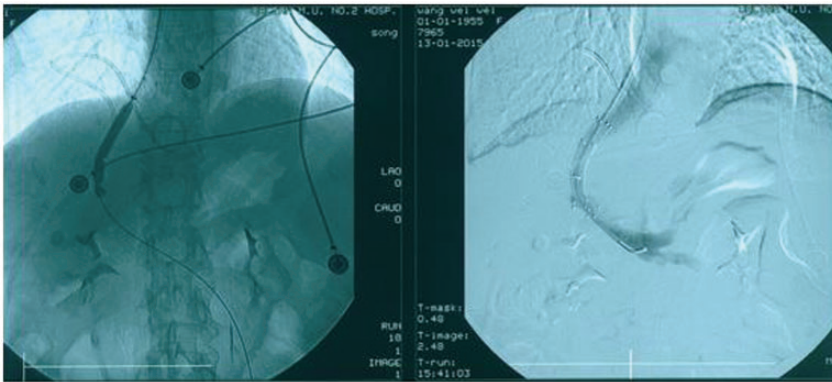
catheter revealed a direct connection between the hepatic and portal veins, and successful puncture of the hepatic vein was achieved. The portal venous pressure was 27 cm H<sub>2</sub>O. Severe varicose hemorrhoids were visualized and embolized with coils (Figure 3). The punctured hepatic vein, portal vein, and parenchyma were expanded with an 8- × 60-mm balloon (Mustang; Boston Scientific, Marlborough, MA, USA),

and coated stents (Fluency; Bard Peripheral Vascular, Tempe, AZ, USA) were deployed (Figure 4). Blood flowed smoothly from the portal vein into the vena cava, and the portal venous pressure was 22 cm H<sub>2</sub>O. Heparin was administered intraoperatively.

Postoperatively, the patient's hematochezia was obviously improved. Her hemoglobin concentration significantly increased



**Figure 3.** Thrombosis in the hemorrhoid venous plexus.



**Figure 4.** Embedded stent.

to 104 g/L. Anticoagulation therapy was administered, and no hepatic encephalopathy occurred; the patient was then discharged.

### Discussion

Portal hypertension causes the development of portosystemic shunts along the gastrointestinal tract, resulting in gastrointestinal varices. Rectal varices arise from communications between the superior hemorrhoidal and middle or inferior hemorrhoidal veins. The incidence of bleeding due to rupture of ectopic varices has increased in recent years.<sup>5</sup> Although rectal variceal bleeding in patients with portal hypertension is

rare, it can be fatal if not appropriately treated.

The prevalence of rectal varices in patients with portal hypertension is increasing due to improved diagnostic techniques, with estimates ranging from 3.6% to 78.0%.<sup>4,6</sup> Diagnosis of rectal varices relies on the findings of endoscopy, angiography, magnetic resonance imaging, computed tomography, and endoscopic ultrasonography. Rectal variceal hemorrhage should be differentiated from hemorrhoidal bleeding because an inaccurate diagnosis can result in fatal blood loss.<sup>7</sup>

Hemorrhoids are vascular cushions comprised of venular and arteriolar anastomoses with no connection to the portal venous

system.<sup>8,9</sup> On proctoscopy they are purple, usually prolapse into the proctoscope, and do not extend proximal to the dentate line. In contrast, anorectal varices are portosystemic collaterals that connect the superior hemorrhoidal vein (portal system) to the middle and inferior hemorrhoidal vein (systemic circulation).<sup>8-10</sup> They are dark blue, extend from the edge of the anus to the rectum, and do not prolapse into the proctoscope. Hemorrhoids are frequently seen in both healthy individuals and patients with portal hypertension,<sup>11,12</sup> and they may coexist with anorectal varices.

There is no consensus on the most appropriate management of rectal variceal rupture, but treatment may be conservative or include endoscopic intervention, surgery, or TIPS placement and angiographic embolization.<sup>6</sup> Interventional therapy, such as injection of tissue adhesive, can successfully control bleeding. Endoscopic ligation is appropriate when the diameter of the varicose vein is less than the caliber of the endoscope. Interventional embolization can result in a short-term satisfactory hemostatic effect, decreasing the rate of bleeding by 94%; however, unless the elevated portal pressure is addressed, most patients will experience rebleeding within 1 year.<sup>13</sup>

Deteriorated liver function is a contraindication to surgery or TIPS placement for management of rectal variceal bleeding. Surgery is usually applicable in patients with Child–Pugh grade A cirrhosis and extrahepatic portal vein thrombosis. TIPS placement involves decompression of the portal venous system to prevent recurrent variceal bleeding. Khaliq et al.<sup>14</sup> first described the use of a TIPS for the treatment of rectal variceal bleeding and indicated that this approach provided a satisfactory hemostatic response and eliminated the possibility of rebleeding. More recently, Ahn et al.<sup>15</sup> described a female patient who was treated with variceal embolization for recurrent rectal variceal

bleeding, which occurred despite successful TIPS placement. Vangeli et al.<sup>16</sup> described 21 patients with cirrhosis who underwent TIPS placement for ectopic variceal bleeding. TIPS was successful in 19 of the 21 patients. In seven patients, TIPS placement and embolization were performed together; of these patients, two developed recurrent bleeding that required further embolization.

We recommend treatment of refractory ectopic variceal bleeding with TIPS placement in combination with embolization. Accordingly, because the patient in the current study developed recurrent rectal variceal bleeding, we implemented TIPS and embolization. Further studies are required to reach a consensus on the most appropriate treatment for rectal variceal bleeding.

#### **Declaration of conflicting interest**

The authors declare that there is no conflict of interest.

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