Successful Treatment of Bleeding Rectal Varices with Balloon-Occluded Retrograde Transvenous Obliteration in a Patient with a History of Sigmoidectomy

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Abstract
Rectal varices most commonly develop in patients with portal hypertension due to liver cirrhosis. Although rectal varices rarely cause bleeding, their rupture can lead to life-threatening massive hemorrhages. Balloon-occluded retrograde obliteration (BRTO) is feasible in cases of varices involving a single or a few drainage vessels. Although rectal varices usually have numerous drainage vessels, the present case involved a single drainage vessel. The unusual presentation of the current case was considered to have been caused by a previous surgery for sigmoid colon carcinoma. BRTO was successfully performed through the left internal iliac vein, achieving complete hemostasis of the rectal varices.

Key words: Rectal varices, Balloon-occluded retrograde obliteration (BRTO), venous intervention, portal hypertension, cirrhosis

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Introduction
The development of portosystemic collateral vessels is common in portal hypertension due to liver cirrhosis [1]. Rectal varices are a type of ectopic varices that occur outside of the gastroesophageal region. Bleeding from rectal varices is rare; however, it has the potential to cause massive life-threatening hemorrhage [2].

Various treatments, such as surgical or endoscopic ligation, endoscopic injection sclerotherapy (EIS), percutaneous transhepatic obliteration (PTO) with modified techniques, transjugular intrahepatic portosystemic shunt (TIPS), and balloon-occluded retrograde transvenous obliteration (BRTO), have been used to control bleeding from rectal varices; however, there is no standard treatment for the condition [2-10]. In general, rectal varices have numerous drainage vessels that pass through the pelvic venous plexus; therefore, BRTO is usually not feasible for rectal varices, owing to their complex hemodynamics.

In this paper, we present a case of a patient with bleeding rectal varices successfully treated with BRTO through a single drainage vein (the left internal iliac vein). We also discuss the treatment options for rectal varices based on their vascular anatomy.

Case Report
A 64-year-old woman with a recurrent tumor (sigmoid colon carcinoma) in her pelvic cavity was admitted to our hospital. She had undergone resection for sigmoid colon carcinoma 4 years prior. She also had liver cirrhosis related to a hepatitis C virus infection and hepatocellular carcinoma (HCC). She had undergone multiple treatments, including partial hepatectomy, radiotherapy, and radiofrequency ablation for HCC during the last 3 years. Radiotherapy was performed for the treatment of recurrent sigmoid colon carcinoma due to poor hepatic functional reserve (Child-Pugh
class C; score: 11). The laboratory tests showed the following results: hemoglobin, 11 g/dL; platelet, 87,000/μL; total bilirubin, 3.5 mg/dL; serum albumin, 2.4 g/dL; and prothrombin time, 61.5%.

Three weeks after the start of radiotherapy, she experienced transient anorectal bleeding twice during the periods of radiation therapy. The contrast-enhanced computed tomography (CT) scan showed large tortuous rectal varices fed by the superior mesenteric vein and draining into the left internal iliac vein (Fig. 1). Three days later, a massive bleeding occurred, and the patient became hemodynamically unstable. Laboratory data showed severe anemia with 7.5 g/dL serum hemoglobin. Emergency endoscopy was performed, and bleeding from large rectal varices was detected (Fig. 2). Although EIS with a mixture of n-butyl-2-cyanoacrylate (histoacryl; B. Braun, Melsungen, Germany) and iodized oil (lipiodol; Andre Guerbet, Aulnay-sous-Bois, France) achieved temporary hemostasis, it was considered that further bleeding could occur. Emergent TIPS creation or PTO might be a therapy of choice; however, these were contraindicated because of marked dilatation of the bile ducts in the left hepatic lobe due to the previous treatment for HCC and multiple peribiliary cysts in the right hepatic lobe, as well as a small amount of ascites. The trans-ileal approach might also be difficult because of adhesion after abdominal surgery. On the basis of this anatomical information, we considered that it might be possible to perform BRTO through the left internal iliac vein. Written informed consent was obtained from the patient, and she was transferred to the angiography room.

Under local anesthesia, a 5-F sheath was first inserted through the right femoral vein. A 0.035-inch hydrophilic guidewire (Radifocus M; Terumo, Tokyo, Japan) was then advanced into the left internal iliac vein through a 5-F shepherd’s hook catheter placed in the left common iliac vein. The catheter and sheath were withdrawn, and a 25-cm-long 7-F sheath that had been steam-shaped into a J-curve was advanced into the left internal iliac vein over the wire. Then, a 6-F balloon catheter (13 mm balloon diameter) (Selecon MP Catheter II; Terumo Clinical Supply, Kakamigahara, Japan) was advanced into the left internal iliac vein. Balloon-occluded venography of the left internal iliac vein showed retrograde opacification of the rectal varices, as well as of the superior and inferior gluteal veins. After embolization of the gluteal veins with microcoils (Nester embolization coil; Cook, Bloomington, IN, USA) through a microcatheter (Progreat β³, Terumo Clinical Supply), stepwise infusion of a sclerosant, 5% EOI (a mixture of 10% ethanolamine olate [Oldamin; Takeda, Osaka, Japan] and the same amount of iopamidol [300 mg I/mL Iopamiron; Bayer, Leverkusen, Germany]), was performed at intervals of 10-30 min through...
a balloon catheter under balloon occlusion. The infusion procedure was ended when the varices had been filled with EOI, and the other collateral vessels of the right internal iliac vein were visualized (Fig. 3). The total volume of infused 5% EOI was 38 mL (stepwise injection of 20, 10, and 8 mL, respectively), and the balloon occlusion period lasted for 70 min. After aspiration of the residual EOI and clots in the vessel lumen through the balloon catheter, the catheter and sheath were withdrawn. During the procedure, 4000 U human haptoglobin (Benesis, Osaka, Japan) was administered intravenously to prevent renal dysfunction. Although 38 mL of EOI was a relatively large volume compared with the patient’s body weight (58 kg), no renal dysfunction developed after BRTO.

After the BRTO procedure, no further bleeding occurred, and the patient’s hemoglobin level stabilized at 11.5 g/dL and the hepatic functional reserve had not been changed. Contrast-enhanced CT scan obtained 2 weeks after the procedure revealed complete obliteration of the rectal varices (Fig. 4). On CT obtained 5 months after BRTO, ascites slightly increased; however, recurrence of the rectal varices and aggravation of other varices were not observed. To date, recurrent rectal bleeding has not developed during the 6-month follow-up period.

**Discussion**

Rectal varices represent portal systemic collaterals that manifest as discrete dilated submucosal veins and constitute a pathway for portal venous flow between the superior rectal veins of the inferior mesenteric system and the middle and inferior rectal veins of the iliac system [1].

The most common cause of rectal varices is portal hypertension due to liver cirrhosis, which is present in 3.6-78% of all cases [2, 3]. Although the clinical incidence of bleeding in patients with rectal varices is 0.5-3.6%, the rupture of rectal varices can lead to massive and life-threatening hemorrhage [2].

The standard treatment for rectal varices is EIS and/or endoscopic variceal ligation [3]. However, when these endoscopic treatments are not sufficient to achieve hemostasis, especially in cases of large varices or massive bleeding, additional therapy might be necessary. Surgical ligation is an effective treatment for bleeding anorectal varices [4]. However, it is not recommended for patients with massive bleeding and/or exhibit severe liver dysfunction. TIPS is also an effective treatment for varices caused by portal hypertension [5-7]. In a report by Vidal et al. [7] on TIPS for the control of ectopic variceal bleeding, hemostasis was achieved in all 24 cirrhotic patients. Rebleeding occurred in six patients, and the cumulative rebleeding rate at 1 and 2 years was 23% and 31%, respectively. PTO can also be used to directly embolize rectal varices. In this procedure, obliteration of the inflow vessel alone enables the eradication of rectal varices, even if they have numerous outflow veins. Additionally, some modifications, such as coil embolization or antegrade balloon occlusion, can enhance the therapeutic effect due to prolonging the retention time of a sclerosant in the varices [8]. However, this procedure is generally contraindicated for patients with ascites. The trans-ileal approach or direct puncturing of the paraumbilical vein is required to access rectal varices in such patients [9].
BRTO is an established procedure for the management of gastric varices in Japan, and has shown promising results in the past decade. The BRTO technique is efficient, especially in cases of varices involving a single or a few drainage vessels. The superior rectal venous plexus normally drains into the portal vein through the inferior mesenteric vein. As portal pressure increases, the superior rectal vein changes from an outflow vessel to an inflow vessel, and drains through the middle and inferior rectal veins into numerous internal iliac branch veins. Although some previous reports have described the use of BRTO to treat rectal varices [2,10], it is not usually indicated for rectal varices because rectal varices usually have multiple drainage veins, and numerous collaterals between the iliac venous branches are often difficult to completely occlude with a balloon catheter. In the present case, the superior rectal vein might have been ligated during the previous sigmoidectomy procedure, and the hemodynamics of the pelvic venous plexus might also have changed. As a result, rectal varices that were drained by a single blood vessel (the left internal iliac vein) developed. On the basis of this specific hemodynamics, BRTO was performed successfully, and complete hemostasis of the rectal varices was achieved. BRTO is considered a less invasive treatment than other interventional procedures such as TIPS and PTO. In cases of rectal varices in which a single drainage vessel is identified on CT, BRTO should be attempted first. Although it is ideal that rectal varices be treated with BRTO alone, the risk of intra procedural rebleeding during occlusion of the outflow should not be ignored. Therefore, a combined technique of BRTO and PTO/TIPS should be considered as an additional option, if possible, especially in high-flow or ruptured cases.

In summary, BRTO is an effective treatment for cases of rectal varices involving a single drainage vein, particularly in patients who had undergone surgical reconstruction in the pelvic cavity.

Conflict of interest: The authors declare that they have no conflicts of interest to report.

References