Treatment of Refractory Stomal Variceal Hemorrhage with Partial Splenic Artery Embolization: A Case Report

Koki Kato¹, Rakuhei Nakama¹, Keiichi Tanimura¹, Masanori Honda¹, Hiroharu Shinozaki¹

Abstract
In this report, we present the case of a patient with recurrent stomal variceal bleeding treated with partial splenic artery embolization (PSE). The patient, a woman in her 60s, had a history of liver cirrhosis and underwent ileocecal resection for ascending colon carcinoma 6 months earlier. The bleeding did not respond to local treatment. Balloon-occluded retrograde transvenous obliteration (BRTO) was performed via the right superficial epigastric vein. However, gross stomal variceal bleeding recurred 2 months post-BRTO. PSE was therefore performed and satisfactory results were obtained. To the best of our knowledge, this is the first case of recurrent hemorrhage from stomal varices that was successfully treated with PSE in a patient with portal hypertension. We consider PSE to be a minimally invasive and definitive treatment for recurrent stomal variceal bleeding.

Key words: Partial Splenic Artery Embolization (PSE), Stomal Variceal Hemorrhage, Portal hypertension

Introduction

Stomal variceal hemorrhage typically occurs in patients with portal hypertension and external enteric diversion. Up to 50% of these patients develop varices, but the associated mortality of stomal hemorrhage is only 3%-4% per bleeding episode compared to the 30%-70% mortality associated with esophageal variceal hemorrhage [1]. Nevertheless, there are significant risks and costs associated with each bleeding episode, which include expenses, inconvenience, and potential morbidity from repeat hospitalizations and blood transfusions. Different treatment regimens with varying degrees of effectiveness have been reported, but reduction of the portosystemic gradient remains the basis of effective long-term treatment [1-6]. Here, we describe a case of a patient with recurrent stomal variceal bleeding that was successfully treated with partial splenic artery embolization (PSE). The procedures described in this article were approved by the review board of our institution and written informed consent was obtained from the patient.

Case Presentation

On multiple occasions, blood was found in the ileostomy bag of a woman in her 60s. She had a history of liver cirrhosis secondary to autoimmune hepatitis. She underwent ileocecal resection for ascending colonic carcinoma 6 months earlier, but an ileostomy was created 2 months after the first surgery because of anastomotic leakage. About 4 months after the creation of the ileostomy, she had several episodes of bleeding from the stoma. Hemostasis was achieved at the first incidence of bleeding by ligating the stomal vessel. However, there was bleeding recurrence 2 weeks later and she was referred for treatment. She had severe hypotension (80/55 mmHg) and a hemoglobin concentration of 7.6 mg/dL despite transfusion. Computed tomography (CT) revealed stomal varices fed by an afferent
The right superficial epigastric vein was accessed and a 5-French (F) introducer sheath was inserted. In brief, a 5.2-F balloon catheter with a maximum balloon diameter of 9 mm (Selecon MP catheter II: Terumo Clinical Supply, Gifu, Japan) was inserted into the right superficial epigastric vein and retrograde venogram was then performed under balloon occlusion. The varices had several collateral veins and a co-axial catheter (Progreat β3, Terumo, Tokyo, Japan) was inserted into the veins, which were then embolized using 5 interlocking detachable coils (3 mm/10 cm × 1, 5 mm/15 cm × 1, and 6 mm/20 cm × 1) (Boston Scientific, Tokyo, Japan) and 16 pushable microcoils (Tornado and Hilal, Cook, Bloomington, IN, USA; and C-Stopper, Piolax, Yokohama, Japan). After confirming the partially visualized stomal varices on balloon-occluded retrograde venogram after coil embolization of the collateral veins (Figure 3), a mixture of 12 mL of 5% ethanolamine oleate (Oldamin, Asuka Pharmaceutical, Tokyo, Japan) and an equal volume of 300 mg/I/mL contrast agent (Iopamiron, Bayer, Osaka, Japan) was injected in the retrograde direction through the right superficial epigastric vein. The balloon was kept inflated for about an hour. After confirming almost total obliteration of the stomal varices with retrograde venography, the catheter was removed.

Contrast-enhanced CT performed 7 weeks post-BRTO showed slight reduction of the varices and incomplete obliteration. There was no variceal bleeding for 2 months post-BRTO and no aggravation of gastroesophageal varices or increased ascites. However, stomal variceal bleeding recurred 2 months post-BRTO with a reduction in hemoglobin concentration. Percutaneous transhepatic obliteration (PTO) was initially considered but we decided to perform PSE because of the thrombocytopenia (50,000/μL) with the expectation of an increase in platelet count and a reduction in portal pressure.
A 6-F guiding sheath (Parent Plus60; Medikit, Tokyo, Japan) was inserted from the right common femoral artery. A selective angiogram of the splenic artery showed a dilated and tortuous splenic artery and an enlarged spleen due to portal hypertension. The venous phase of splenic arteriography showed venous reflux to the inferior mesenteric vein (Figure 4). A coaxial catheter (Progreat β3, Terumo, Tokyo, Japan) was inserted into the lower and middle branches of the splenic artery, and embolization was then performed using gelatin sponge (Spongel; Astellas Pharma, Tokyo, Japan) particles and a microcoil (Tornado embolization coil with a diameter of 6 mm, Cook). The gelatin sponge particles were suspended in a solution of 0.25 g of the antibiotic meropenem hydrate (Meropenem; Sumitomo Dainippon Pharma, Osaka, Japan) mixed with a nonionic iodine contrast agent (Iomeron 350; Eisai, Tokyo, Japan). Angiography of the splenic artery performed after embolization showed occlusion of the distal branches except for those at the upper pole of the spleen. The venous phase of the angiogram of the splenic artery performed after embolization showed ablated peripheral splenic parenchyma and no reflux into the inferior mesenteric vein (Figure 5). We reckoned that this indicated decompression of the portal pressure. Several days after the procedure, the patient complained of a slight fever and left upper abdominal pain, which were treated using only medications. The platelet count tripled 1 month after the procedure and contrast-enhanced CT performed 2 months post-PSE showed reduced stomal varices (Figure 6). The embolized volume of the spleen determined from contrast-enhanced CT was 74%.

The patient had no recurrence of bleeding and no progression of gastroesophageal varices. Liver function tests were normal. There were no incidences of post-procedural complications, which include worsening ascites, and there were no incidences of stomal bleeding in 3 years of follow-up.

Discussion

There are various management strategies for stomal variceal bleeding. Although local procedures (such as local pressure application, recumbent body positioning, and surgical suturing and ligation) are effective for initial bleeding control, they may be ineffective for the prevention of recurrent bleeding and may have limited long-term efficacy [1, 2]. Surgical revision is not recommended for many patients on account of cirrhosis and portal hypertension. A complete revision of the stoma requires laparotomy with its attendant risks and complications. Portocaval shunting to reduce portal hypertension is effective for the prevention of recurrent bleeding and has been used successfully; however, it also carries similar perioperative risks and risks of recurrent
bleeding and encephalopathy [2]. It is also associated with significant morbidity and mortality, which ranges from 5% to 15% [2].

Anecdotally, there are 2 pathologic conditions that cause recurrent stomal variceal bleeding. Some stomas are diffusely congested and bleed diffusely, while others bleed from a focal site, such as a mesenteric varix. Patients with stomas that are diffusely congested or engorged with diffuse venous blood have more favorable outcomes after transjugular intrahepatic portosystemic shunt (TIPS) decompression. Placement of a TIPS is an effective alternative for treating stomal variceal bleeding; however, it may sometimes result in secondary hepatic encephalopathy or liver failure in patients with decompensated liver function [3].

Bleeding from focal varices in the stoma (with the rest of the stomal mucosa looking normal and not engorged) can be treated with TIPS placement (if the portal vein, the mesenteric vein, or both are patent) or with transvenous obliteration. BRTO, PTO, and trans-TIPS balloon-occluded antegrade transvenous obliteration can all be suitable approaches of transvenous obliteration [3]. In the case presented here, gross stomal variceal bleeding recurred 2 months post-BRTO via the right superficial epigastric vein. The degree of transvenous obliteration may have been insufficient; however, it may have prevented stomal damage. In contrast, Yata et al. reported a case of a patient with stomal variceal bleeding treated with sclerotherapy via a retrograde approach that developed massive ascites after the procedure [4]. They maintained that reducing blood flow to the stoma may be enough for patients with a short prognosis.

There are several reports on the direct percutaneous approach [5]. This approach reduces procedural time, is simple, highly effective, and the least invasive approach. Naidu et al. embolized afferent veins with metallic coils after inserting a catheter into the vessel through punctured stomal varices [5]. In our patient, it was possible to directly puncture the stomal varices through the mucosa but we decided against it because of the risk of infection.

Kishimoto et al. reported successful treatment of patients with stomal varices using PTO [6]. PTO is less invasive than TIPS placement and other surgical procedures. The potential complications of PTO include bleeding, bile leakage, liver trauma, and portal thrombosis [2]. In fact, PTO was initially planned as the second treatment for our patient but we decided to perform PSE because of the thrombocytopenia. However, embolization of the mesenteric vein using metallic coils usually has no long-term effect, and re-bleeding may occur from newly developed varices as a result of persistent portal hypertension. As a result, subsequent embolization is made even more difficult. Furthermore, Spier et al. reported that sclerotherapy carries a risk of stomal damage [1]. In our patient, stomal variceal bleeding recurred 2 months post-BRTO. Although incomplete embolization of variceal veins may lead to re-bleeding, it may also prevent stomal damage.

PSE is used to improve liver function, reduce variceal bleeding, treat hepatic encephalopathy, and improve blood cell counts in patients with portal hypertension [7]. In addition, PSE can increase platelet count in patients with hypersplenism from idiopathic thrombocytopenic purpura, thalassemia, idiopathic hypersplenism, and cytopenia caused by anticancer chemotherapy [7-10]. The improved liver function observed in patients with cirrhosis following PSE is likely related to increased blood flow in the hepatic arterial and superior mesenteric vein [8]. PSE can arrest variceal bleeding as a result of decreased splenic venous flow into the portal venous system, leading to reduction in mesenteric and colonic venous pressures. Helaly et al. investigated the acute effects of PSE on portal and splanchic hemodynamics in patients with cirrhosis using duplex examinations. They concluded that PSE has an immediate portal decompression effect on patients with portal hypertension without reducing portal flow [8].

However, since PSE may be unable to stop massive stomal variceal bleeding, local procedures (such as local pressure application, surgical suturing, and ligation) should be tried first. These local procedures have limited long-term efficacy. We consider PSE to be an important strategy for the prevention of recurrent stomal variceal bleeding.

Splenic abscess is the most critical complication of PSE. There are several possible mechanisms of splenic infection and they include: anaerobic bacterial growth in the hypoxic tissue, percutaneous contamination with exogenous bacteria, and retrograde transport of enteric pathogens via reversed portal flow [9]. Several studies have reported that a large splenic infarction volume and a large infarction ratio are obvious risk factors for complications after PSE [7]. However, the effects of prophylactic intra-arterial antibiotic agents have never been discussed. Masada et al. [9] reported that the use of intra-arterial antibiotic agents may be of benefit in the prevention of splenic inflammation after PSE.

The long-term efficacy of PSE is still unclear. The study by Palsson et al. has the longest follow-up time of all relevant studies and included a cohort of 26 patients, whose ongoing therapy for over 20 years was standard and consisted of pharmacological treatment (beta-blockers and diuretics) and repeat endoscopic sclerotherapy treatment of esophageal varices; however, this was found to be insufficient and the patients had to be treated with PSE [10]. In this patient group, the number of bleeding episodes from esophageal varices was significantly reduced after PSE. The efficacy of PSE may stretch over a longer period than expected.

In conclusion, our results show that PSE can be useful for the control of stomal variceal bleeding, but more studies may be necessary to further confirm the effectiveness and safety of this procedure in the treatment of stomal varices.

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

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