

CASE REPORT

원위부 십이지장 정맥류의 대량 출혈에 대한 풍선-폐쇄 역행 경정맥 폐쇄술의 장기적인 치료 성공 1예

황순우, 손주현, 김태엽, 김지연, 이지영, 광동신, 김해수, 송순영¹

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Long-term Successful Treatment of Massive Distal Duodenal Variceal Bleeding with Balloon-occluded Retrograde Transvenous Obliteration

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Duodenal variceal bleeding in patients with portal hypertension due to cirrhosis or other causes is uncommon. We report on a case of a 55-year-old male with an ectopic variceal rupture at the distal fourth part of the duodenum who presented with massive hematochezia and shock. Shortly after achievement of hemodynamic stability, due to the limitation of an endoscopic procedure, we initially attempted to find the bleeding focus by abdominal computed tomography, which showed tortuous duodenal varices that drained into the left gonadal vein. He was treated with first-line balloon-occluded retrograde transvenous obliteration (BRTO), resulting in a favorable long-term outcome without rebleeding three years later. This case suggests that BRTO may be a first-line therapeutic option for control of ruptured duodenal varices, especially at a distal location. (**Korean J Gastroenterol 2014;63:248-252**)

Key Words: Bleeding, duodenal; Varix, ectopic; Cirrhosis; Treatment; Balloon occlusion

INTRODUCTION

Duodenal varices are an unusual cause of gastrointestinal hemorrhage in patients with portal hypertension due to cirrhosis or other causes, and variceal bleeding from the fourth part of duodenum is rare. Management of duodenal varices is difficult due to the potential for massive bleeding and limitations of an endoscopic approach. Its treatment may require use of a multidisciplinary team approach involving hepatologists, endoscopists, interventional radiologists, and surgeons.¹ There is a paucity of information regarding guidelines

for treatment of duodenal varices. However, the optimal therapeutic modality depends mainly on the site of the varices, patient's condition, doctor's experience, and the usefulness of equipment.

In performance of first-line endoscopy, subjects with suspected ectopic variceal bleeding should be in a hemodynamically stable state. If endoscopic treatment fails, interventional radiological techniques, such as embolization, transjugular intrahepatic portosystemic shunt (TIPS), or balloon-occluded retrograde transvenous obliteration (BRTO) should be the next step to consider.¹

Received July 28, 2013. Revised September 17, 2013. Accepted September 18, 2013.

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Financial support: None. Conflict of interest: None.

BRTO is a new therapeutic technique used mainly for control of life-threatening bleeding from large gastric varices using a splenorenal shunt.² In recent years, BRTO has been applied for the treatment of duodenal variceal bleeding, and successful results have been reported.³⁻⁷ However, these reports are limited to patients with failed endoscopic therapy in the second portion of the duodenum. If a patient's condition is unstable and the endoscopic device cannot reach the bleeding site, the clinician is faced with difficulty in control of bleeding. In addition, there is no consensus regarding the best method for control of duodenal variceal bleeding.⁸ Here, we report on a long-term successful treatment using first-line BRTO in a hemodynamically unstable patient with massive variceal bleeding at the fourth part of the duodenum.

CASE REPORT

A 55-year-old man was admitted to the emergency room with massive hematochezia and syncope, which had started 5 hours previously. He had a history of a small amount of intermittent hematochezia with hemorrhoids for one year. The patient had a history of hypertension without medication for four years. He had no history of viral hepatitis, bleeding disorders, jaundice, or blood transfusion. He had a 20-year daily drinking average of 80 g of alcohol. The patient complained of vomiting without hematemesis. On admission, his blood pressure was 80/50 mmHg, pulse rate was 110/min, and body temperature was 36.7°C. He appeared to be acutely ill and had an alert mental status. His conjunctiva was anemic (very pale) and his tongue was mildly dehydrated. Abdominal examination revealed distension with no tenderness. Laboratory data on admission were as follows: hemoglobin, 4.6

g/dL; mean corpuscular volume, 70.2 fl; hematocrit, 15.8%; white blood cell, 15,400/ μ L with 84.6% neutrophil; platelet, 167,000/ μ L; prothrombin time, 20.5 seconds (INR 2.02); serum total protein, 5.3 g/dL; albumin, 2.6 g/dL; total bilirubin, 1.5 g/dL; ALT, 170 U/L; AST, 56 U/L; GGT, 538 U/L. The results of serologic testing for hepatitis B and hepatitis C were negative.

One hour after admission, his blood pressure was 70/40 mmHg and his mental status was drowsy. He sustained a symptom of hematochezia and we initially treated him with fluid resuscitation with normal saline, blood, and antibiotic prophylaxis. Due to his hypotension with mental deterioration, we did not attempt an emergency upper endoscopy. The patient was transferred to the intensive care unit and ventilator care was administered. After empiric administration of terlipressin and achievement of hemodynamic stabilization, to search for the bleeding focus, we performed an abdominal CT scan, which showed tortuous ectopic varices in the fourth part of the duodenum; these drained into the left gonadal vein (Fig. 1). Based on the results of the imaging study and the difficulty of an endoscopic approach, BRTO was planned first.

With the patient in a supine position, a 9-French guiding catheter (TFE Transjugular catheter; Cook Co., Bloomington, IN, USA) was inserted into the inferior vena cava via the right internal jugular vein. An 8-mm occlusion balloon catheter (Boston Scientific, Natick, MA, USA) was inserted into the left gonadal vein through the left renal vein. Finally, we selected the dilated efferent vein of the duodenal varices. We performed retrograde venography with balloon inflation; however, contrast filling into the duodenal varices failed due to multiple collateral variceal draining veins. After successful emboli-

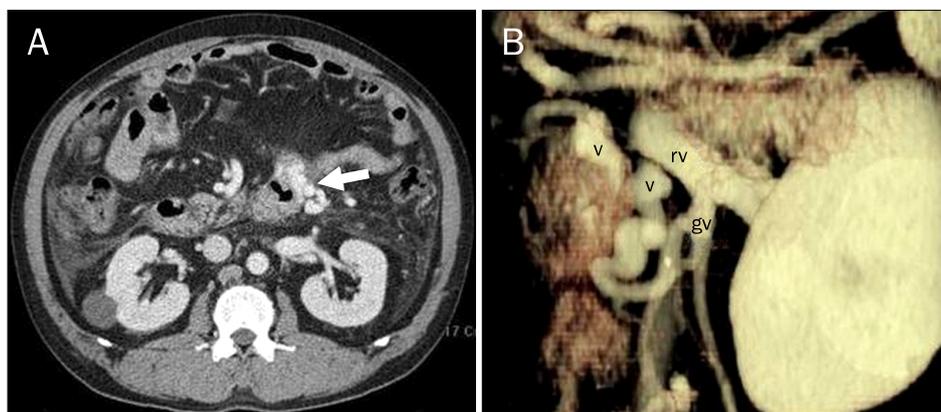


Fig. 1. (A) Axial images of a portal-phase abdominal CT scan show varices (arrow) of the fourth part of the duodenum. (B) These duodenal varices (v) drained into the left renal vein (rv) through the left gonadal vein (gv).

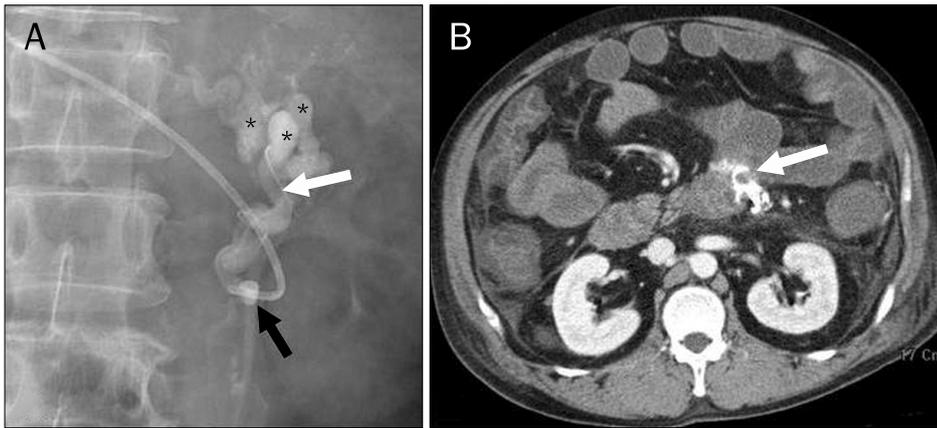


Fig. 2. (A) Radiograph obtained after infusion of a mixture of a sclerotic agent and lipiodol through a microcatheter (white arrow) during balloon occlusion (black arrow) shows duodenal varices and the afferent vein (asterisks). (B) Axial image of a portal phased abdominal CT shows filling of the sclerosant within duodenal varices by the sclerosant (arrow).

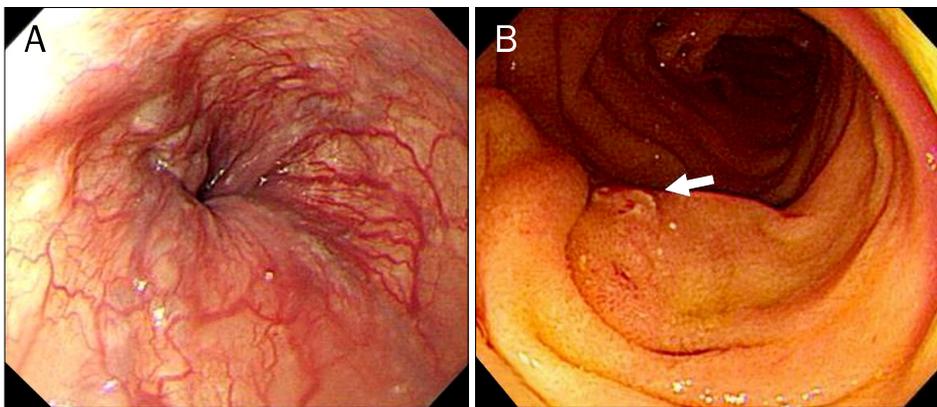


Fig. 3. (A) Two weeks after the balloon-occluded retrograde transvenous obliteration procedure, gastroscopy showed small esophageal varices at the lower esophagus. (B) Push enteroscopy showed ruptured erosion (arrow) on the varix at the fourth part of the duodenum.

zation of the collaterals with the infusion of gelatin sponge particles, a 3-French microcatheter (Progreat; Terumo, Tokyo, Japan) was advanced into the duodenal varices. During balloon occlusion, a sclerosing agent, a mixture of 5% ethanolamine oleate and lipiodol (5 : 1), was infused through the microcatheter. Infusion was continued until the duodenal varices and an afferent vein were imaged (Fig. 2A). The occlusion balloon was maintained for 30 minutes. An abdominal CT image obtained immediately after the procedure confirmed filling of the variceal lumen with the sclerosant (Fig. 2B). After the procedure, his vital signs were stable and his mental status improved.

Two weeks later, an upper endoscopy was performed, which showed small esophageal varices and portal hypertensive gastropathy. Push enteroscopy showed duodenal varices with recently ruptured erosion at the fourth portion of the duodenum (Fig. 3).

His hepatic venous pressure gradient was measured as 27 mmHg. He was managed with diuretics, non-selective beta-blocker, rifaximin, and lactulose; the patient then made a

full recovery. One month after BRTO, he was discharged without procedure related complication. The patient had no recurrence of bleeding three years after BRTO and no evidence of recurrence of duodenal varices was found on a follow-up CT scan performed three years later.

DISCUSSION

Duodenal variceal bleeding in patients with portal hypertension due to cirrhosis or other causes is uncommon.⁹ Although various procedures for control of duodenal variceal hemorrhage have been previously reported, those reports were limited to bleeding of the second portion of the duodenum, which was treated by standard upper endoscopy,^{5-7,10-13} while only one case of hemorrhage from the lower duodenum, where endoscopy could not be inserted, was treated with BRTO.⁸ In this case, we successfully performed first-line BRTO due to a hemodynamically unstable situation and location of varices in the far distal duodenum detected by CT.

Among the duodenal varices, duodenal bulb is the most

common site, followed by the second portion.¹⁴ In our case, duodenal varices located in the fourth part of the duodenum and variceal bleeding from the fourth part of the duodenum is much rarer than from the bulb or the second portion of the duodenum. Duodenal varices tend to have a smaller diameter and shorter length than esophageal varices, and they are located deeper and mainly closer to the serosa.¹⁵ Therefore, the risk of bleeding from the duodenal varices is relatively low and no therapeutic modality has been established, mainly due to its rarity.⁸ However, because the blood flow of duodenal varices is rapid, the initial presenting manifestation of duodenal varices is often massive bleeding by ruptured erosion, resulting in a life-threatening condition, as in our patient.^{16,17}

In the first-line investigation for all patients with suspected variceal bleeding, endoscopy should be performed as soon as possible. If endoscopy fails to identify the source of bleeding, video capsule endoscopy, technetium TC-99m red blood cell scintigraphy, CT angiography, or other modalities should be the next step in detection. Due to hemodynamic instability, an endoscopic procedure was not suitable in our patient. Therefore, we performed an abdominal CT scan, and ectopic varices were then detected in the fourth part of the duodenum.

Various methods have been tried for management of duodenal variceal bleeding, including endoscopic procedures (band ligation, sclerotherapy), embolization via an interventional radiological technique, surgical treatment, TIPS, and BRTO.¹ However, no standard guideline has been established because no randomized studies on this condition have been reported, only limited case reports or small case series.⁸ Therefore, management depends on the location of the varices, possibility of an endoscopic approach, patient's condition, availability of therapeutic modalities, and the clinician's experience.

Endoscopic therapies include band ligation, vascular obstruction, and injection sclerotherapy. Band ligation is a useful method for treatment of reachable duodenal varices, as shown in a few reports; however, it can be unsafe if varices are too large to exceed endoscopy diameter^{9,10} because of the risk of bleeding from mucosal defects developed after ligation. Therefore, first-line band ligation followed by BRTO is beneficial in such a situation.¹⁶ Injection sclerotherapy has been used for achievement of hemostasis in patients with esophageal or gastric variceal bleeding with various

sclerosants. It can also be applied to duodenal variceal bleeding and we previously reported on a case of successful control of duodenal variceal bleeding with cyanoacrylate injection at the second portion of the duodenum.¹³ However, complications of injection sclerotherapy using cyanoacrylate can be serious, especially for duodenal varices, such as systemic embolization, bowel perforation, and recurrent bleeding, because, due to the duodenal anatomy, determining the extent of the varices and maintaining full visualization of the lesions is difficult. In addition, the thinness of the duodenal wall, compared to esophagus and stomach, can increase the risk of perforation and complications of the endoscopic procedure.

Interventional radiological modalities, such as TIPS and BRTO, are the next steps for patients with duodenal variceal hemorrhage in whom endoscopic treatment has failed.⁹ TIPS is a useful method for control of refractory bleeding¹²; however, there are potential risks of encephalopathy, aggravated liver dysfunction, and procedure-related complications.¹⁸

BRTO has been accepted as a therapeutic modality for large gastric varices, and is considered valuable for patients with hepatic encephalopathy or portal vein thrombosis. However, due to low availability and technical difficulties, its use is limited.

In the current case, we first performed BRTO before endoscopic treatment. This decision was justified by the fact that the patient had decompensated cirrhosis, hemodynamic instability, the location of bleeding vessel on CT, and experienced practitioners.

Recently, Tanaka summarized 12 cases of duodenal variceal bleeding successfully treated with BRTO.⁸ Hashimoto et al.¹⁷ reported on a case in which cyanoacrylate, not ethanolamine oleate, was used first as a sclerosant during BRTO for duodenal varices. In all of these cases and in our case, complete hemostasis was achieved and rebleeding did not occur during the various follow-up periods. The reported complication during BRTO for duodenal varices is induction of esophageal varices.⁷ Occlusion of the duodenal varices and dilated draining veins might have induced the esophageal varices.⁸ However, BRTO offers several benefits in treatment of duodenal varices. First, the procedure is safe and effective. Second, it can directly eliminate intravariceal blood flow. Third, BRTO can reach varices in anatomically complex locations. Finally, BRTO can be used safely in patients with poor liver function

and even promotes improvement of the Child-Pugh score.¹⁹

In conclusion, bleeding from ectopic varices must be considered for patients with chronic liver disease and hematochezia. This case suggests that BRTO is a safe and effective hemostatic procedure for the management of ruptured duodenal varices, especially in the distal portion, which cannot be reached by endoscopic treatment.

REFERENCES

1. Helmy A, Al Kahtani K, Al Fadda M. Updates in the pathogenesis, diagnosis and management of ectopic varices. *Hepatol Int* 2008;2:322-334.
2. Kanagawa H, Mima S, Kouyama H, Gotoh K, Uchida T, Okuda K. Treatment of gastric fundal varices by balloon-occluded retrograde transvenous obliteration. *J Gastroenterol Hepatol* 1996; 11:51-58.
3. Ohta M, Yasumori K, Saku M, Saitsu H, Muranaka T, Yoshida K. Successful treatment of bleeding duodenal varices by balloon-occluded retrograde transvenous obliteration: a transjugular venous approach. *Surgery* 1999;126:581-583.
4. Tominaga K, Montani A, Kuga T, et al. Combined balloon-occluded embolization for treatment of concurrent duodenal, gastric, and esophageal varices: a case report. *Gastrointest Endosc* 2001;53:665-668.
5. Ota K, Okazaki M, Higashihara H, et al. Combination of transileocolic vein obliteration and balloon-occluded retrograde transvenous obliteration is effective for ruptured duodenal varices. *J Gastroenterol* 1999;34:694-699.
6. Takamura K, Miyake H, Mori H, et al. Balloon occluded retrograde transvenous obliteration and percutaneous transhepatic obliteration for ruptured duodenal varices after operation for rectal cancer with multiple liver metastasis: report of a case. *J Med Invest* 2005;52:212-217.
7. Zamora CA, Sugimoto K, Tsurusaki M, et al. Endovascular obliteration of bleeding duodenal varices in patients with liver cirrhosis. *Eur Radiol* 2006;16:73-79.
8. Tanaka O, Ohno K, Ohno T, et al. Should balloon-occluded retrograde transvenous obliteration be the first-line interventional radiologic treatment for bleeding duodenal varices? A case report and review of the literature. *Acta Radiol* 2008;49:32-36.
9. Norton ID, Andrews JC, Kamath PS. Management of ectopic varices. *Hepatology* 1998;28:1154-1158.
10. Fayad N, Nammour F, Elfant A. Endoscopic variceal ligation for bleeding duodenal varices. *J Clin Gastroenterol* 2004;38:467.
11. Barbish AW, Ehrinpreis MN. Successful endoscopic injection sclerotherapy of a bleeding duodenal varix. *Am J Gastroenterol* 1993;88:90-92.
12. Almeida JR, Trevisan L, Guerrazzi F, et al. Bleeding duodenal varices successfully treated with TIPS. *Dig Dis Sci* 2006;51: 1738-1741.
13. Son BK, Sohn JH, Chang MH, Park YK, Kim TY, Jeon YC. A case of successful endoscopic injection sclerotherapy with N-butyl-2-cyanoacrylate for ruptured duodenal varices. *Korean J Gastroenterol* 2007;49:336-340.
14. Amin R, Alexis R, Korzis J. Fatal ruptured duodenal varix: a case report and review of literature. *Am J Gastroenterol* 1985;80: 13-18.
15. Park SB, Lee SH, Kim JH, et al. Successful treatment of duodenal variceal bleeding by endoscopic clipping. *Clin Endosc* 2013; 46:403-406.
16. Akazawa Y, Murata I, Yamao T, et al. Successful management of bleeding duodenal varices by endoscopic variceal ligation and balloon-occluded retrograde transvenous obliteration. *Gastrointest Endosc* 2003;58:794-797.
17. Hashimoto R, Sofue K, Takeuchi Y, Shibamoto K, Arai Y. Successful balloon-occluded retrograde transvenous obliteration for bleeding duodenal varices using cyanoacrylate. *World J Gastroenterol* 2013;19:951-954.
18. Poncic RJ, Kowdley KV. Paradoxical cerebral emboli after transjugular intrahepatic portosystemic shunt and coil embolization for treatment of duodenal varices. *Am J Gastroenterol* 1997; 92:1372-1373.
19. Koito K, Namieno T, Nagakawa T, Morita K. Balloon-occluded retrograde transvenous obliteration for gastric varices with gastrosplenic or gastrocaval collaterals. *AJR Am J Roentgenol* 1996; 167:1317-1320.