

Case Report

A case of ruptured duodenal varices treated successfully by endoscopic injection sclerotherapy under radiographic guidance with a mixture of N-butyl-2-cyanoacrylate-lipiodol

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Abstract

While guidelines have been established for the treatment of esophageal and gastric varices, there is as yet no consensus with regard to the optimal method of treatment of ectopic varices. We report a case of ruptured duodenal varices treated successfully by endoscopic injection sclerotherapy (EIS) under radiographic guidance with a mixture of N-butyl-2-cyanoacrylate (NBCA)-lipiodol. A 73-year-old female patient with a history of liver cirrhosis was transferred to our hospital for the treatment of ruptured duodenal varices. Esophagogastroduodenoscopy (EGD) revealed distended duodenal varices with a mucosal erosion and a coagulum in the descending portion of the duodenum. Dynamic contrast-enhanced computed tomography (CT) of the abdomen revealed duodenal varices arising from the pancreaticoduodenal vein, being a branch of the superior mesenteric vein, the efferent vessel draining through the veins of Retzius. A total of 6 mL of an NBCA + lipiodol mixture (mixing ratio, 3:1) was injected into 3 sites of the duodenal varices via a 23-gauge disposable injection needle. Radiographic imaging during the EIS demonstrated that the NBCA + lipiodol mixture had reached the distended veins connected to the duodenal varices. At the follow-up CT conducted 6 days after the EIS, the duodenal varices with its collateral vessels were almost completely obliterated by the NBCA + lipiodol mixture. Even a single EIS session eradicated the varices almost completely without any complications. EIS under radiographic guidance using a mixture of NBCA-lipiodol (75%NBCA) is an effective method of treatment of duodenal varices in hemorrhagic or elective cases.

(Key words: duodenal varices, endoscopic injection sclerotherapy, N-butyl-2-cyanoacrylate, lipiodol)

Introduction

Duodenal varices, which are ectopic varices, are rare as compared to esophagogastric varices. Duodenal varices are formed by the development of collateral vessels in the presence of portal hypertension. There has been an increasing number of reports of duodenal varices in recent years. Bleeding from duodenal varices is usually massive and often fatal. There is no established method of treatment for ruptured duodenal varices. We report a case of ruptured duodenal varices that was treated successfully by endoscopic injection sclerotherapy (EIS) under radiographic guidance using a mixture of N-butyl-2-cyanoacrylate (NBCA) -lipiodol .

Case report

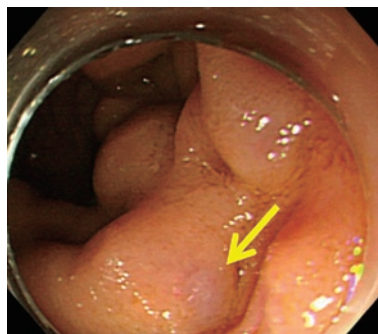
A 73-year-old female patient was admitted to a local hospital with a 7-day history of melena. She had been under regular follow-up for non-B, non-C, non-alcoholic liver cirrhosis (but no hepatocellular carcinoma) for 2 years. She gave no previous history of gastrointestinal hemorrhage or treatment for esophageal varices. The hemoglobin level measured at admission was 5.7 g/dL. Esophagogastroduodenoscopy (EGD) was performed on the third hospital day, which revealed distended duodenal varices with a mucosal erosion and a coagulum in the descending portion of the duodenum (Figure 1). Although esophageal varices (Li, F₁, Cb, RC₀) were also detected, there were no obvious bleeding points, gastric varices, or portal hypertension-associated gastropathy. The patient was transfused with 8 units of packed red blood cells and referred to our hospital on the fourth hospital day for the treatment of ruptured duodenal varices. At arrival at our hospital, physical examination revealed that the patient was fully conscious and alert, and afebrile, with a heart rate of 75 beats/minute and blood pressure of 106/68 mmHg. The patient was anemic, but had no jaundice, lymphadenopathy or edema. Abdominal examination revealed a soft enlarged spleen palpable 6 cm below the left costal margin, but no signs of ascites. The laboratory data at admission are summarized in Table 1.

Figure 1: Endoscopic findings

- Esophagogastroduodenoscopy (EGD) revealed distended duodenal varices in the descending portion of the duodenum.
- A mucosal erosion (arrow) on the distended duodenal varices was the bleeding source.



a)



b)

The Child-Pugh grade was assessed as B. Hemoglobin was 7.1 g/dL (normal range, 11.3-15.2), the serum levels of tumor markers (AFP and PIVKAlI) were within normal limits. Dynamic contrast-enhanced computed tomography (CT) of the abdomen revealed duodenal varices arising from the pancreaticoduodenal vein, being branches of the superior mesenteric vein, the efferent vessel draining through the veins of Retzius (Figure 2). We have assumed that the efferent vessel was the distended left ovarian vein through the veins of Retzius. But it was difficult to clearly demonstrate the continuity of the gonadal vein and the veins of Retzius by multidetector-row CT (Abdominal angiography was not performed). Because of the high risk of re-bleeding as assessed from the endoscopic appearance (distended duodenal varices with a mucosal erosion), EIS was performed for the ruptured duodenal varices. A total of 6 mL of an NBCA + lipiodol mixture (mixing ratio, 3:1) was injected into 3 sites of the duodenal varices via a 23-gauge disposable injection needle. A 50% glucose solution was administered intravenously before and after the injection of the NBCA + lipiodol mixture, as in the case of ordinary endoscopic treatment for gastric varices. Radiographic imaging during EIS demonstrated that the NBCA + lipiodol mixture reached the distended veins connected to the duodenal varices (Figure 3). At the follow-up CT performed 6 days after the EIS, the duodenal varices with the collateral vessels were almost completely obliterated by the NBCA + lipiodol mixture (Figure 4). Furthermore, EGD, also performed 6 days after the EIS, revealed active ulcers on the duodenal varices, but no evidence of bleeding (Figure 5). The patient's postoperative recovery was uneventful and she was discharged home in a stable condition. At the follow-up CT performed 3 months after EIS, further decrease in the size of the duodenal varices was observed.

Table 1: Laboratory data on admission

WBC	5260 / μ l	TP	4.4 g/dl
Hb	7.1 g/dl	Alb	2.3 g/dl
Ht	21.6 %	T-Bil	1.05 mg/dl
Plt	16.4x10 ⁴ / μ l	AST	37 IU/L
		ALT	32 IU/L
prothrombin time	92%	LDH	133IU/L
		γ GTP	78IU/L
AFP	3.7 ng/ml	BUN	27 mg/dl
PIVKAlI	12 mAU/ml	Cr	0.77 mg/dl
		CRP	1.68 mg/dl
HBsAg(-)	HBcAb(-)	NH3	33 μ g/dl
HCVAb(-)			

Figure 2: Dynamic contrast-enhanced CT of the abdomen revealed duodenal varices (arrow 1) arising from the pancreaticoduodenal vein (arrow 2), being a branch of the superior mesenteric vein, the efferent vessel (arrow 3) draining through the veins of Retzius.

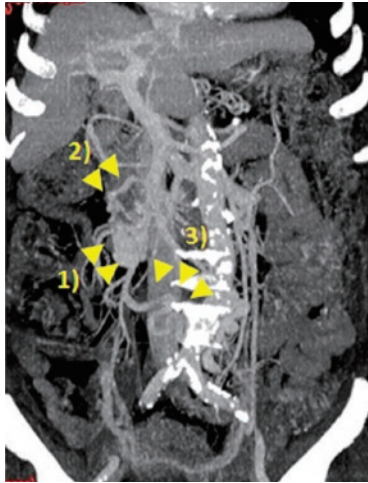
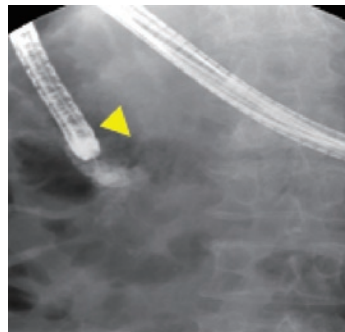


Figure 3: Radiographic imaging during EIS

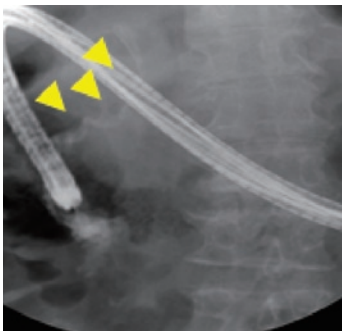
- NBCA-lipiodol appearing in the duodenal varices.
- NBCA-lipiodol coming into view in the efferent vessel (arrow).
- NBCA-lipiodol appearing in the afferent vessel (the pancreaticoduodenal vein) (arrow).
- NBCA-lipiodol being injected into another site of the duodenal varices.



a)



b)



c)



d)

Figure 4: At follow-up CT performed 6 days after the EIS, the duodenal varices with the collateral vessels were almost completely obliterated by the NBCA-lipiodol. duodenal varices (arrow 1), pancreaticoduodenal vein (arrow 2), efferent vessel draining through the veins of Retzius (arrow 3).

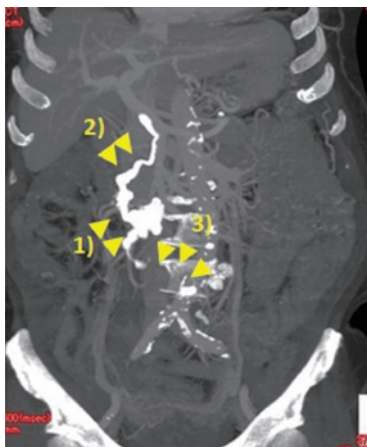


Figure 5: Six days after the EIS, EGD revealed active ulcers on the duodenal varices, but no bleeding.



Discussion

Ectopic varices are relatively rare; however, in the United States and Europe, approximately 5% of all varices associated with gastrointestinal bleeding are identified as ectopic varices^{1,2)}. A nationwide questionnaire survey conducted in 1990 showed that ectopic varices were diagnosed at an extremely low frequency in Japan (129/18,540 cases; 0.7%), with the lesion sites varying widely, and including the duodenum, small intestine, colon, rectum, and gallbladder³⁾. A study of 173 cases of ectopic varices by Watanabe et al. in 2010 revealed that rectal varices accounted for 44.5% of all cases of ectopic varices, duodenal varices for 32.9%, small intestinal varices for 6.4%, colonic varices for 3.5%, and other varices for 12.7%⁴⁾. The descending portion of the duodenum is the most common site of duodenal varices in Japan, whereas the duodenal bulb is reported as the most common site overseas. This discrepancy between Japan and Western countries is considered to be related to the differences in the causative disorders: in Western countries, liver cirrhosis accounts for only about 30% of the cases, while extrahepatic portal vein obstruction with splenic vein obstruction or superior mesenteric vein obstruction due to thrombo-

sis, tumor or pancreatitis are the more common causes; on the other hand, liver cirrhosis is the most frequently reported causal disorder (80.3%) of ectopic varices in Japan⁴. In liver cirrhosis and posthepatic portal hypertension, the pancreaticoduodenal vein and jejunal vein, being branches of the superior mesenteric vein, show hepatofugal blood flow to form varices, mostly in the descending or transverse parts of the duodenum⁴. It is of importance to delineate the variceal hemodynamic profiles in order to understand the pathophysiology of ectopic varices and establish suitable therapies; MR angiography, multidetector-row CT and CT angiography are essential imaging examinations for this purpose^{5,6}.

Matsui et al. reported that the hemorrhagic factor in duodenal varices is the F factor, and not the RC sign⁷. Guidelines have been established for the treatment of esophageal and gastric varices⁸, whereas there is still no consensus with regard to the most suitable treatment method for ectopic varices. While the propriety of prophylactic treatment of ectopic varices is still a subject of debate, it is agreed some treatment should be undertaken for hemorrhagic or elective cases.

Bleeding from ectopic varices may be fatal because the rapid and abundant blood flow can make it difficult to achieve hemostasis. Endoscopic therapy is often performed in emergent cases, whereas B-RTO is performed in elective and prophylactic cases. Balloon-occluded retrograde transvenous obliteration (B-RTO) is practicable for the treatment of duodenal varices, because the inferior pancreaticoduodenal vein constitutes the afferent vessel and the ovarian or testicular vein comprises the efferent vessel in about half of the cases of duodenal varices. In Japan, B-RTO is frequently performed for the treatment of duodenal varices after primary hemostasis, by either EIS using NBCA or endoscopic variceal ligation in cases with bleeding⁴. It is generally thought to be preferable to provide complete occlusion of the varices by interventional radiologic therapies, such as B-RTO and percutaneous transhepatic obliteration when the afferent and efferent vessels are delineated in elective and prophylactic cases^{4,9-14}.

In our hemorrhagic case, we detected by multidetector-row CT that the afferent vessel was the pancreaticoduodenal vein, being a branch of the superior mesenteric vein, and that the efferent vessel drained through the veins of Retzius. We then performed EIS under radiographic guidance for the ruptured duodenal varices, using a mixture of NBCA + lipiodol (75%NBCA). Endoscopic oblitative therapy with NBCA is useful for emergency control of acute gastric and ectopic variceal bleeding. NBCA polymerizes immediately upon contact with blood, resulting in rapid hemostasis. It is now the treatment of first choice worldwide for obliteration of bleeding gastric varices^{15,16}. Lipiodol prevents the tissue adhesive from polymerizing too quickly and also allows for radiographic monitoring. The mixing ratio for NBCA-lipiodol presents a problem with respect to embolism. When the proportion of lipiodol is increased to obtain a better contrast effect, the NBCA becomes too dilute and does not polymerize well at the injection site, increasing the risk of embolism. Fluoroscopic observation with infusion of 75% NBCA (while avoiding flow into the systemic circulation) was performed to determine the extent of the varices. Even during the initial phase of endoscopic varicealography during injection sclerotherapy (EVIS), the NBCA + lipiodol mixture appeared in the duodenal varices. Then, the NBCA-lipiodol was visualized in the efferent vessel. Finally we terminated the injection when we observed its appearance in the afferent vessel (the pancreaticoduodenal vein).

Complications of EIS using NBCA for varices include fever and pain due to an inflammatory response. Major complications include ulceration and recurrent bleeding, although more serious complications, including embolization to the brain, portal vein, lung, and spleen have also been reported. Other compli-

cations include bacteremia, abscesses, and development of visceral fistula¹⁷⁻²¹. To minimize the risk of embolic complications, we recommend that endoscopists use the smallest volume of NBCA-lipiodol necessary for obliteration, under radiographic guidance.

By radiographically guided EVIS, a single EIS session eradicated the varices almost completely without any complications. Ideally, that should indicate about changes over time endoscopic findings of duodenal varices after the EIS. But all of follow up EGD were not performed due to circumstances of the patient. Therefore, CT findings alone, the distinction of duodenal varices in the duodenal wall or wall outside were difficult. EIS under radiographic guidance using a mixture of NBCA + lipiodol (75%NBCA) is an effective method for the treatment of duodenal varices in hemorrhagic or elective cases.

Although EIS using NBCA is currently the treatment of first choice worldwide for obliteration of bleeding gastric varices, EIS using NBCA is not officially approved or covered by the National Health Insurance in Japan. Therefore, informed consent from the patients is necessary.

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十二指腸静脈瘤破裂に対し透視下に実施した lipiodol と n-butyl-2-cyanoacrylate の混合液による内視鏡的硬化療法が有用であった1例

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池谷 敬, 東海 浩一, 池田 正俊, 牛丸 信也,
浅野 岳春, 松本 吏弘, 岩城 孝明, 福西 昌徳,
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要 約

食道・胃静脈瘤の治療指針はほぼ確立されているが、異所性静脈瘤の治療法については一定の見解が得られていない。我々は、十二指腸静脈瘤破裂に対して、透視下に行った内視鏡的硬化療法が有用であった1例を経験した。症例は73歳、肝硬変の女性で十二指腸静脈瘤破裂にて当院へ紹介となった。内視鏡検査にて、十二指腸下行脚に出血点と思われるびらんを伴うF3の静脈瘤を認めた。腹部CTでは脾十二指腸静脈を供血路としRetzius静脈に排血路を伴う十二指腸静脈瘤を認めた。n-butyl-2-

cyanoacrylate と lipiodol を3:1に混合し、X線透視下に確認しながら3ヶ所に計6.0ml 静脈瘤内に供血路、排血路が造影されるまで局注した。6日後の造影CTでは、静脈瘤から連続する供血路と排血路の一部に lipiodol の集積を認め、静脈瘤はほぼ硬化剤により置換されていた。透視下に硬化剤の注入範囲を確認しながら内視鏡的硬化療法を行うことで合併症なく、また追加治療を必要としない十分な十二指腸静脈瘤の治療が可能であった。