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Abstract

The term “ectopic varices” is used to describe dilated portosystemic collateral veins in unusual locations other than the gastroesophageal region. We recently experienced a rare case of ectopic varices that developed in the gastroduodenal anastomosis after subtotal gastrectomy. A 70-year-old male with liver cirrhosis due to hepatitis C virus infection was admitted for hematemesis and tarry stool. He had received a subtotal gastrectomy with the Billroth-I method for gastric ulcer at 46 years of age. Although emergency endoscopy revealed esophageal and gastric fundal varices, there were no obvious bleeding points. After removal of the coagula, ectopic varices and a fibrin plug were observed on the gastroduodenal anastomosis. During the observation, blood began to spurt from the fibrin plug. N-butyl-2-cyanoacrylate with lipiodol injection succeeded in hemostasis. Splenic angiography showed gastric varices feeding from a short gastric vein and the posterior gastric vein. The blood flow around the bleeding point, as indicated by lipiodol deposition, had decreased, and no feeding vein was observed. Endoscopic and angiographic findings are shown and the treatment for such lesions is discussed.

KEYWORDS: ectopic varices, N-butyl-2-cyanoacrylate (Histoacryl), gastroduodenal anastomosis, portal hypertension

Case Report

Ectopic Varices Rupture in the Gastroduodenal Anastomosis Successfully Treated with N-butyl-2-cyanoacrylate Injection

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The term "ectopic varices" is used to describe dilated portosystemic collateral veins in unusual locations other than the gastroesophageal region. We recently experienced a rare case of ectopic varices that developed in the gastroduodenal anastomosis after subtotal gastrectomy. A 70-year-old male with liver cirrhosis due to hepatitis C virus infection was admitted for hematemesis and tarry stool. He had received a subtotal gastrectomy with the Billroth-I method for gastric ulcer at 46 years of age. Although emergency endoscopy revealed esophageal and gastric fundal varices, there were no obvious bleeding points. After removal of the coagula, ectopic varices and a fibrin plug were observed on the gastroduodenal anastomosis. During the observation, blood began to spurt from the fibrin plug. N-butyl-2-cyanoacrylate with lipiodol injection succeeded in hemostasis. Splenic angiography showed gastric varices feeding from a short gastric vein and the posterior gastric vein. The blood flow around the bleeding point, as indicated by lipiodol deposition, had decreased, and no feeding vein was observed. Endoscopic and angiographic findings are shown and the treatment for such lesions is discussed.

Key words: ectopic varices, N-butyl-2-cyanoacrylate (Histoacryl), gastroduodenal anastomosis, portal hypertension

Ectopic or aberrant varices are dilated portosystemic venous collaterals at unusual locations, and they are typically associated with portal hypertension. Ectopic varices have been reported to develop in various organs such as the duodenum, colon, gall bladder, uterus, urinary bladder and abdominal sto-

mas [1-8]. However, varices other than gastroesophageal or rectal are rare entities [3]. We experienced here a rupture of ectopic varices located in the gastroduodenal anastomosis which was successfully treated with N-butyl-2-cyanoacrylate (Histoacryl) injection. Endoscopic and angiographic findings are shown and the treatment for such lesions is discussed.

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Case Report

A 70 year-old male was admitted for hematemesis and tarry stool. He had received a subtotal gastrectomy with the Billroth-I method for gastric ulcer at 46 years of age. He received blood transfusion at that time and suffered from liver cirrhosis due to hepatitis C virus infection. Laboratory data on admission were follows: hemoglobin 7.7 g/dl, white blood cell $11,300/\mu\text{l}$, platelet $10.1 \times 10^4/\mu\text{l}$, total protein 5.7 g/dl, Alb 2.5 g/dl, T-Bil 0.6 mg/dl, AST 46 IU/l, ALT 66 IU/l, γ -GTP 69 U/l, BUN 37 mg/dl, Cr 0.8 mg/dl, and prothrombin time 53%. The hepatitis B antigen was negative and the hepatitis C antibody was positive. Blood pressure was 88/52 mmHg. Although emergency endoscopy revealed esophageal and gastric fundal varices, there were no obvious bleeding points (Fig. 1). There were no ulcer findings in the stomach. Removal of the coagula revealed ectopic varices and a fibrin plug on the gastroduodenal anastomosis (Fig. 2A). During the observation, blood began to spurt from the fibrin plug (Fig. 2B). A total of 0.8 mL, N-butyl-2-cyanoacrylate (Histoacryl) with lipiodol (mixed ratio, 1:0.6) injected using a 23-gauge disposal injection needle succeeded in inducing hemostasis (Fig. 2C). Twenty percent glucose was infused before and after N-butyl-2-cyanoacrylate with lipiodol injection, as in the ordinary endoscopic treatment for gastric varices. After

hemostasis, the color of the collateral vein in the stomach wall (Fig. 2D) and esophageal varices (Fig. 2E) changed to blue.

Abdominal X-ray after endoscopic hemostasis showed lipiodol deposition near the Petz clamp from the gastrectomy. Splenic angiography showed gastric varices feeding from a short gastric vein and the posterior gastric vein (Fig. 3). The left gastric vein was not shown on angiography. It might have collapsed due to ligation during the subtotal gastrectomy or to N-butyl-2-cyanoacrylate injection. There was no obvious spleno-renal shunt. The blood flow around the bleeding point, as indicated by lipiodol deposition, had decreased and no feeding vein was observed at this time (Fig. 3). The endoscopic findings at 1 month from treatment showed N-butyl-2-cyanoacrylate (Histoacryl) with lipiodol deposition and no recurrence of varices (Fig. 2F).

Discussion

The recognition of varices at unusual sites has long been described in the literature, since Alberti *et al.* [9] described duodenal varices in 1931. Ectopic varices have been reported in the duodenum, ileum, cecum, ascending, descending and rectosigmoid colon, gall bladder, uterus, vagina, urinary bladder and abdominal stomas [2-10]. Standard diagnostic and therapeutic procedures have not yet been established

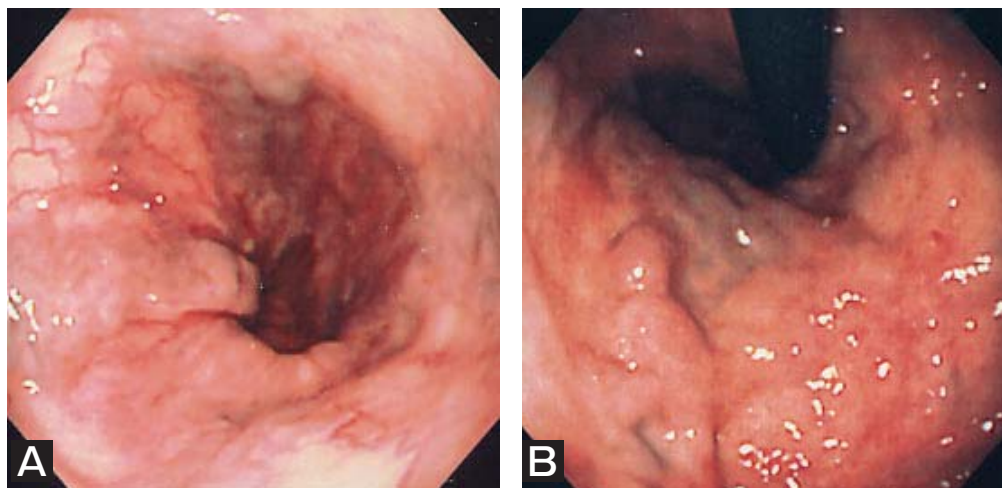


Fig. 1 Emergency endoscopy revealed esophageal (A) and gastric fundal varices (B). There was no obvious bleeding from these varices. There were no ulcer findings in the stomach.

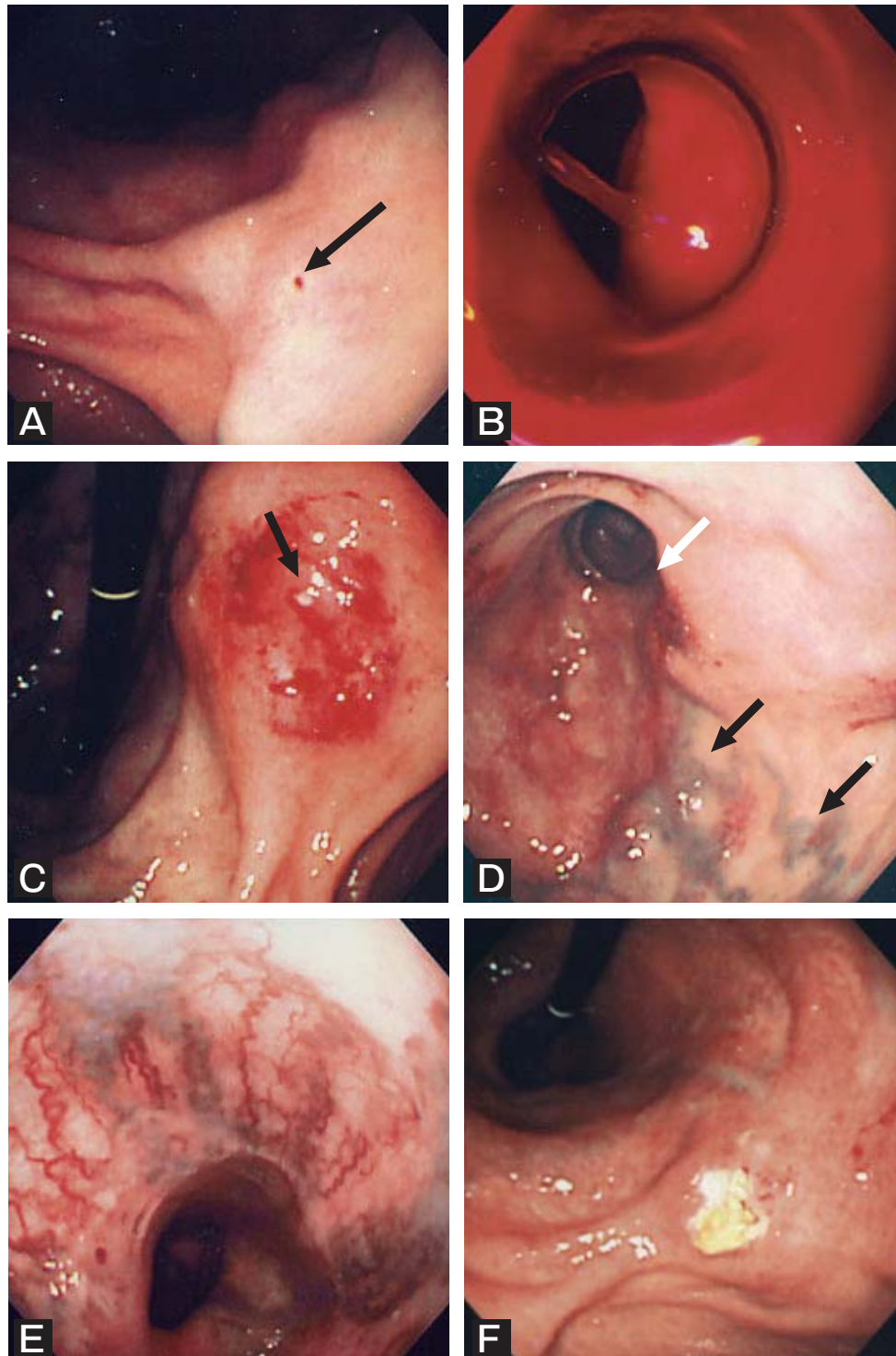


Fig. 2 Endoscopic findings of ectopic varices in the gastroduodenal anastomosis. **A**, After removing the coagula, ectopic varices and a fibrin plug on the gastroduodenal anastomosis were revealed; **B**, During the observation, blood began to spurt from the fibrin plug; **C**, N-butyl-2-cyanoacrylate (Histoacryl) with lipiodol injection succeeded in inducing hemostasis; **D**, **E**, After hemostasis, the color of the collateral vein in the stomach wall (**D**, black arrow) and esophageal varices (**E**) changed to blue. White arrow; bleeding point; **F**, After 1 month from treatment, N-butyl-2-cyanoacrylate (Histoacryl) with lipiodol deposition was observed, with no recurrence of varices.

for ectopic intestinal varices, which are rarely formed on other parts of the digestive tract than the gastroesophageal region [11]. Although several promising treatments have been reported, bleeding ectopic varices are regarded as potentially life-threatening [12]. Regarding ectopic varices after gastrectomy, several cases with esophagojejunal varices after total gastrectomy have been reported [13–16]. However, varices developing on the gastroduodenal anastomosis as in our case are rare. Of all patients with gastrointestinal varices in Japan, those with ectopic varices after gastrectomy are only 0.06% (12/18,540) [17, 18]. Eleven of these 12 cases were ectopic varices after total gastrectomy using the Roux-Y method [17].

The management of ectopic varices is frequently difficult and controversial. Bleeding from ectopic varices is rare and accounts for only between 1% and 5% of all variceal bleeding [3]. However, once the bleeding starts, it becomes difficult to control and is sometimes fatal. Chikamori *et al.* [14, 15] reported that percutaneous transhepatic obliteration (PTO) is a useful technique to treat bleeding esophagojejunal

varices after total gastrectomy. In our case, N-butyl-2-cyanoacrylate (Histoacryl) with lipiodol injection was useful in achieving hemostasis. A surgical resection or interventional embolization of varices is sometimes useful when the varices are localized. However, surgical options, such as a portosystemic shunt or variceal ligation, are limited to selected patients. Unfortunately, many patients are not good operative candidates for such treatment modalities. Somatostatin analog or β -blocker has also been used to control bleeding from varices [3]. The transjugular intrahepatic portosystemic shunt (TIPS) procedure is an effective modality in the therapy of cirrhotic patients with bleeding from ectopic varices unresponsive to conservative management [19–21]. However, there are data showing that TIPS frequently fails to eradicate gastric varices; the reported success rate is only 50% [22]. Moreover, there is recent evidence showing a higher re-bleeding rate after creation of a TIPS, compared to trans-catheter sclerotherapy [23].

In our hospital, treatment strategies for emergency bleeding from varices are as follows: esophageal varices receive endoscopic variceal ligation (EVL); gastric varices receive N-butyl-2-cyanoacrylate injection or endoscopic clipping. After hemostasis with emergency treatment, additional treatment strategies are as follows: esophageal varices receive endoscopic injection sclerotherapy (EIS), gastric varices receive interventional radiology (IVR) such as balloon-occluded retrograde transvenous obliteration (BRTO) or PTO.

In conclusion, we experienced a rare case of a rupture of ectopic varices located on the gastroduodenal anastomosis. The patient was successfully treated with N-butyl-2-cyanoacrylate injection, which proved very useful in this case.



Fig. 3 Splenic angiography showed gastric varices feeding from a short gastric vein (black arrow) and the posterior gastric vein (white arrow). There was no obvious spleno-renal shunt. The blood flow around the bleeding point, as indicated by lipiodol deposition (arrow head), had decreased, and no feeding vein was observed at this point.

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