Case Report

Unusual Bleeding From Hepaticojejunostomy Controlled by Adult Variable Stiffness Colonoscopy: Report of a Case and Literature Review

Hiroyuki Baba^{1,2}, Mai Wakabayashi², Atsushi Oba², Hironobu Baba², Akito Mitsuoka², Hiroshi Nakamura², Takahiro Sanada², Hiroshi Kuwabara², Kazumi Nakajima², Narihide Goseki², Hideyuki Ishida¹

¹*Saitama Medical Center, Saitama Medical University, Saitama, Japan*

²*Shuwa General Hospital, Kasukabe, Japan*

We herein present a case of a 59-year-old man who had undergone pylorus preserving pancreaticoduodenectomy with regional lymph node dissection prior to episodes of melena. Series of conventional endoscopic investigations failed to identify the bleeding source. Enhanced computed tomography scan revealed complete obstruction of the main portal vein with numerous collateral veins running towards the hepatic hilus. Comprehensively, hemorrhage from the jejunal varices caused by postoperative portal hypertension was highly suspected. As the jejunal loop was out of reach, adult variable-stiffness colonoscope (AVSC) was utilized to solve the Roux-en-Y anatomy. Numerous telangiectasis and small varices at hepaticojejunostomy were observed and in the mean time, bleeding was noticed and endoclips were placed without any delay. Ectopic variceal bleeding in jejunal loop after pancreaticoduodenectomy is difficult to manage. We believe that AVSC is an alternative device when specialized jejunal endoscopy is not available.

Key words: Hepaticojejunostomy – Adult variable stiffness colonoscopy – Endoclip – Jejunal varicose vein – Portal hypertension

Reprint requests: Hiroyuki Baba, MD, PhD, Department of Digestive Tract and General Surgery, Saitama Medical Center, Saitama Medical University, 1981 Kamoda, Kawagoe, Saitama, 350-8550, Japan.
Tel.: +81 49 228 3619; Fax: +81 49 222 8865; E-mail: hirobaba@saitama-med.ac.jp

Bleeding ectopic varices in the jejunal loop after biliary reconstruction is a rare entity.¹ Management of such bleeding is difficult and is not well defined. Here, we report a case of a pancreatic cancer patient who had pylorus-preserving pancreaticoduodenectomy (PD) 2.5 years prior to variceal bleeding at hepaticojejunostomy (HJ) anastomosis (Roux-en-Y fashion). We successfully treated this out-of-reach afferent jejunal loop bleeding by using adult variable stiffness colonoscopy (AVSC).

Case Report

A 59-year-old man experienced occasional dyspnea and melena during his follow-up after surgery. He had a history of pancreatic head cancer [T3N2M0 Stage IV, Union for International Cancer Control (UICC) classification, 2010] diagnosed 2.5 years ago and had undergone pylorus-preserving PD with regional lymph node dissection. Postoperative course was uneventful, and adjuvant chemotherapy was carried out using gemcitabine for 1 year. His performance status (PS) was well kept (PS, 0–1). Experiencing somewhat stronger fatigue than usual, he visited our emergency room and was admitted to our hospital for diagnosis and treatment.

On admission, his blood pressure was below 100 mmHg (systolic) and tachycardia was present. His hemoglobin was 4.8 g/dL, so he was given 4 units of red blood cell transfusion and intravenous fluids to stabilize his hemodynamics. Emergency upper gastrointestinal (GI) endoscopy was carried out but failed to detect any bleeding responsible for melena. As anticipated, we were unable to access the afferent limb of the Roux-en-Y anastomosis because the length of the endoscopy was too short. Computed tomography (CT) scan revealed complete obstruction of portal blood flow, causing portal hypertension, and numerous cavernous vessels at the hepatic hilus were observed (Fig. 1). Retroperitoneal recurrence of the pancreatic cancer was detected. We were convinced that hemorrhage was a result of the rupture of jejunal varicose vein formed at the afferent loop of Roux-en-Y anatomy.

Because conventional gastroduodenoscopy had failed to reach HJ previously, we diverted to AVSC. Inside the jejunal limb, we noticed massive red wine-colored fluid. Irregular spider web-like veins and varix were observed at HJ anastomosis (Fig. 2A). During the observation, passive bleeding was noticed (Fig. 2B), and endoclip was applied immediately (Fig. 2C). Within the next few days, melena had stopped and his hemodynamic status was

Fig. 1 Irregular collateral veins at hepatic hilus.

stabilized. As a long-term treatment principle, his hemoglobin level was kept low on purpose, just below the normal limits, with the intention to avoid further bleeding at least by keeping the blood pressure relatively low. He was discharged after 10 days of admission. Fortunately, on 10-month follow-up, no bleeding was observed. Irregular vessels were visible, but varicose vein had disappeared (Fig. 2D).

Discussion

In this case report, we have demonstrated the necessity of investigating the afferent jejunal loop in patients with a history of PD when unexpected melena occurs. Source of bleeding may include not only ectopic varices, but also pseudoaneurysms and ulcers. Nevertheless, endoscopic approach is ideal for direct diagnosis and immediate treatment. However, anatomic difficulty prevents us from reaching the jejunal afferent loop with conventional gastroduodenoscopy. Therefore, longer endoscopy is necessary and, in practice, we have clearly shown that AVSC can be a choice, and universal hemostat procedures using endoclips are reliable.

Recent reports have confirmed the accessibility of single-balloon endoscopy (SBE)^{2–4} and double-balloon endoscopy (DBE)^{5–8} for Roux-en-Y biliary anastomosis. Compared with conventional gastroduodenoscopy, SBE and DBE are longer and have the advantage of anchoring the endoscope by using the balloon overtube to insert more deeply. SBE and DBE seem to be equally useful for diagnostic and therapeutic procedures.^{9,10} However, these special-

Fig. 2 Endoscopic findings of the hepaticojejunostomy. (A) Varicose vein and irregular formation of small vessels. (B) During observation, bleeding occurred unexpectedly. (C) Endoclip was applied immediately. (D) Ten-month follow-up.

ized devices are not always available when necessary. Ironically, Akahoshi *et al* reported the advantages of DBE only if it is available upon treatment.⁸

Literature discussing the use of AVSC for upper GI investigation and treatment, particularly for Roux-en-Y anatomy, are rare. Chahal *et al* have reported the usefulness of AVSC for endoscopic retrograde cholangiography in post-orthotopic liver transplant population with Roux-en-Y biliary anastomosis.¹¹ Azeem *et al* compared AVSC, SBE and pediatric colonoscope and concluded SBE is a little better in procedural success than AVSC.¹² AVSC is available in most institutions now, and it can be an alternative device for SBE and DBE. However, their structures are not designed for upper GI investigation; because the diameter of the endoscopy is larger and the overall stiffness is much more than the conventional gastroduodenoscopy, it may be a stress for the patient's throat. But still, we cannot ignore its

length and end-viewing character. Furthermore, it enables us to use commercial hemostat devices.

Reviewing the previously reported literature of jejunal variceal rupture at HJ, portal hypertension was caused by either recurrence of cancer or adhesion (Table 1). Among the diseases listed, cancer was cited most often, 10 times including our case. Other causes were chronic pancreatitis, bile duct stones, acute cholecystitis, choledochojejunostomy cyst, and familial intrahepatic cholestasis needing liver transplantation. Various treatments have been carried out through the years: portal vein dilatation, portal stenting, bilioenteric reanastomosis, embolization, splenectomy, shunt operation, and endoscopic procedures.^{11–14,17–26}

Neumann *et al*¹³ first reported the treatment using endoscopy. Their case was also a patient with pancreatic cancer whose portal vein was obstructed 1 year after operation. They first tried to use

Table 1 Published papers of varicose vein rupture at hepatico- or choledochojejunostomy anastomosis

Patient No.	Author	Age/sex	Primary disease	Operation	Complication	Duration from initial operation	Treatment	Year
1	Stafford Johnson ¹⁷	39/M	Chronic pancreatitis	Cystadenectomy and CJ	None	Unknown	Portal vein dilatation and stenting	1997
2	Ishida ¹⁸	75/F	Gallbladder cancer	CJ	None	16 months	Reanastomosis	1998
3	Hiraoka ¹⁹	66/F	Pancreatic cancer	PD with IORT	None	1 year	Portal vein dilatation and stenting	2001
4	Hiraoka ¹⁹	44/F	Cholelithiasis	Cholecystectomy and CJ	None	16 years	Portal vein dilatation and stenting	2001
5	Sato ²⁰	79/F	Gallbladder cancer	Cholecystectomy and CJ	Leakage	7/9 years	Resection of jejunum and embolization	2003
6	Sakai ²¹	54/F	Bile duct cancer	PD, portal vein reconstruction, and IORT	None	16 months	Portal vein dilatation and stenting	2005
7	Ota ²²	64/M	Cancer of ampulla of Vater	PpPD	Cholangitis	8 years	Portal vein dilatation and stenting	2005
8	Smith ²³	42/F	Chronic pancreatitis	CJ, gastrojejunostomy, and cholecystectomy	None	18 months	Ligation of dilate jejunal vein/repair of CJ	2005
9	Taniguchi ²⁴	64/F	Hilar cholangiocarcinoma	Right hepatic tommy and caudate lobe tommy	None	5/10 years	Reanastomosis and splenectomy	2008
10	Rezende-Neto ²⁵	64/F	Acute cholecystitis	CJ	None	15 years	Splenectomy and shunt operation	2008
11	Neumann ¹³	78/F	Pancreatic cancer	PpPD	None	1 year	Endoscopic therapy	2008
12	Sasamoto ²⁶	42/M	Intrahepatic and choledochojejunostomy stones	CJ	Leakage	1 year	Embolization	2010
13	Curcio ¹²	17/F	Familial intrahepatic cholestasis	Split liver transplantation	None	15 years	Endoscopic therapy	2010
14	Prachayakul ¹¹	29/F	Choledochal cyst	Choledochal cyst excision	None	6 months	Endoscopic therapy	2011
15	Saeki ¹⁴	59/M	Osteoclastic giant cell tumor of the duodenum	PD	None	3 years	Embolization	2012
16	Saeki ¹⁴	79/F	Hilar cholangiocarcinoma	Right hepatic tommy and caudate lobe tommy	Cholangitis and portal vein thrombosis	13 years	Endoscopic therapy and shunt operation	2012
17	Present case	59/M	Pancreatic cancer	PpPD	None	30 months	Endoscopic therapy	2013

F, female; IORT, Intraoperative Radiation Therapy; M, male; PpPD, pylorus-preserving pancreaticoduodenectomy.

colonoscopy but failed to reach the afferent loop, so they switched to DBE. Argon plasma laser was used for hemostat.¹³ Prachayakul *et al* used SBE and injected histoacryl into the varicose vein, and although successfully done, thrombosis at the branch of superior mesenteric vein occurred.¹⁴ Saeki *et al* used DBE and applied Aethoxysklerol (Kaigen Pharma Co., Ltd., Osaka, Japan) for sclerotherapy, but rebleeding occurred 1 week later, forcing them to perform shunt operation.¹⁵ Curcio *et al* used SBE and endoclips for hemostat, although the clip placement caused varix rupture and cyanoacrylate injection was carried out.¹⁶ To our knowledge, we are the first to utilize AVSC for ectopic jejunal variceal bleeding in Roux-en-Y anatomy. We believe that we can manage this out-of-reach bleeding in a practical manner that can be done in most places, without regard to the presence of advanced endoscopies. To date, only sporadic case reports are available; we need to accumulate more patients to define what the best hemostat procedure might be.

Our present case did not experience episodes of rebleeding subsequently. We were surprised to find that his treatment was carried out only once, despite the etiology being similar to rupture of esophageal varix. Looking at the treatment course from all sides, a few reasons may be hypothesized. First, hemodynamics of portal blood flow may have changed during the course. Although the evaluation of portal blood flow was not carried out to confirm the change, newly formed esophageal varix on follow-up gastroduodenoscopy may support this hypothesis. Second, keeping the overall blood pressure low by controlling the hemoglobin level just below the normal limits and refraining from excess intravenous infusion of extracellular fluids may have helped to lower the portal blood pressure, which may have kept telangiectasis from becoming worse.

Conclusion

Ectopic varicose vein at jejunal afferent loop should be considered when unidentified GI tract hemorrhage is experienced in patients with Roux-en-Y anatomy. AVSC can be an alternative for SBE or DBE by means of length and hemostat procedure. Endoclip seems to be a better technique since sclerosant injection may give rise to unwanted thrombosis.

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