Clinical Case Report

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Unusual bleeding from hepaticojejunostomy controlled by side-to-side splenorenal shunt

A case report

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Abstract

Rationale: Ectopic variceal bleeding due to hepaticojejunostomy (HJ) is unusual and difficult to manage. Reports on the use of side-to-side splenorenal shunt for severe bleeding from varices at HJ anastomosis are lacking.

Patient concerns: A 43-year-old man was admitted to our hospital with repeated episodes of hematemesis. He has a history of right hemihepatectomy with HJ reconstruction to the left hepatic duct for hilar cholangiocarcinoma. Two years after surgery, he presented with repeated episodes of hematemesis and underwent blood transfusion.

Diagnoses: Imaging tests and endoscopic investigation failed to identify the bleeding source. When conservative management failed to control his bleeding, he underwent emergency laparotomy, which revealed hemorrhage from ectopic varices at the HJ anastomosis.

Interventions: To arrest the bleeding, a side-to-side venovenal anastomosis was created between the splenic and left renal veins to form a shunt for decompression of the varices at the HJ anastomosis.

Outcomes: After the surgery, the patient's symptoms ceased, and a no bleeding in the digestive tract was noted at 2-year follow-up.

Lessons: The present patient is the first reported case of unusual bleeding from HJ controlled by a side-to-side splenorenal shunt. We believe this approach is a useful and effective surgical treatment for severe bleeding from varices at the HJ anastomosis.

Abbreviations: DSA = digital subtraction angiography, GI = gastrointestinal, HJ = hepaticojejunostomy, MDCT = multidetector computed tomography.

Keywords: hepaticojejunostomy, portal hypertention, splenorenal shunt, upper gastrointestinal bleeding

1. Introduction

Although hemorrhage of upper gastric and intestinal varices is a common complication of portal hypertension,[1,2] ectopic variceal bleeding due to hepaticojejunostomy (HJ) is uncommon, which is largely due to portal vein stenosis or obstruction. To date, only 15 such cases have been reported in English literature. Hence, the management of this condition remains difficult and unclear. Here, we report a case of a patient with hilar cholangiocarcinoma who had undergone right hemihepatectomy 2 years prior to variceal bleeding at the HJ anastomosis. We successfully treated this uncontrolled bleeding using a side-to-side splenorenal shunt.

2. Case report

A 43-year-old man with a 2-year history of repeated episodes of hematemesis was admitted to our hospital. Three years before his presentation, he underwent right hemihepatectomy with reconstruction HJ of the left hepatic duct for Klatskin tumor (hilar cholangiocarcinoma). During the operation, the left branch of the portal vein was injured and immediately repaired. The patient treated with analgesia, hemostasis, nutritional support, use of expectorants and antibiotics but without anticoagulants after surgery. Postoperatively on day 3, he underwent re-laparotomy for thrombectomy of the left main portal vein thrombosis. Because of liver insufficiency secondary to reduced hepatic inflow due to thrombosis, he developed massive intra-abdominal bleeding and recurrent portal vein thrombosis for which he underwent further laparotomy to arrest hemorrhage and thrombectomy. He finally made a full recovery and was discharged 49 days after admission. He was doing well until 2 years later when he presented with repeated episodes of hematemesis requiring hospital admission for upper and lower gastrointestinal (GI) endoscopy followed by blood transfusion.

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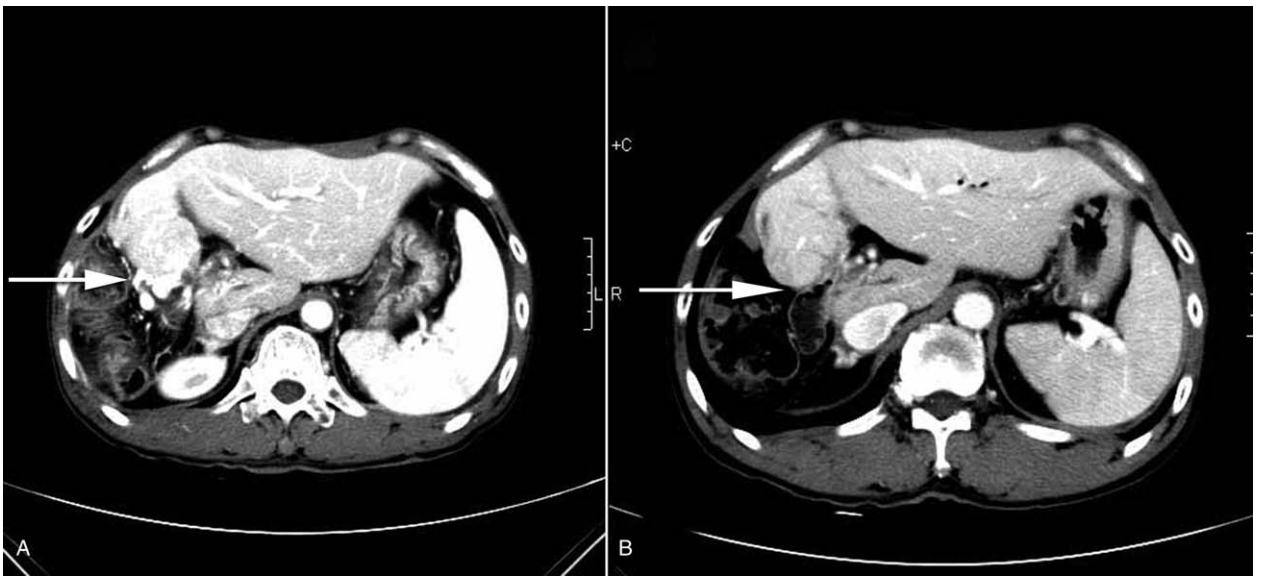

Figure 1. The varicose vein disappeared after the operation. (A) Preoperative MDCT. White arrow, varicose vein. (B) Postoperative MDCT. White arrow, non-varicose vein. MDCT = multidetector computed tomography.

However, the intervals between the 2 episodes became shorter and each bout became more severe. Endoscopy failed to reveal any varices in the esophagus, stomach, colon, or rectum. Multidetector computed tomography (MDCT) with angiography revealed obstruction and cavernous transformation of the portal vein with multiple collaterals and varices at the site of the HJ anastomosis. Digital subtraction angiography (DSA) failed to identify the source of hemorrhage on 2 separate occasions when he was actively bleeding. Technetium-99m-labeled red cell scintigraphy also failed to show the source of the bleeding. Eventually, he underwent emergency laparotomy as a life-saving measure when conservative management failed to arrest his bleeding on his latest admission. During laparotomy, there was no free blood in the abdominal cavity, but a grossly dilated afferent jejunum was found. After division of extensive adhesions, avoiding bleeding from multiple collaterals, the HJ anastomosis was finally exposed and examined. To arrest the bleeding, a side-to-side venovenal anastomosis was created between the splenic and left renal veins to form a shunt to decompress the varices at the HJ anastomosis. The operation time lasted 351 minutes, and he had blood loss of 3500 mL, necessitating transfusion of 16 units of red cell suspension with 8 units of fresh frozen plasma. Postoperatively, there was no further bleeding. Clinically, he became more stable and was discharged 21 days later. One month later, MDCT revealed no collaterals or varices at the site of the HJ anastomosis (Figs. 1–3). No bleeding of the digestive tract was note at 2-year follow-up.

3. Discussion

Ectopic varices are uncommon, accounting for <5% of cases of variceal bleeding.[2,3] Diagnosis of ectopic variceal bleeding due to HJ is always difficult because of its low incidence and atypical clinical presentation. Thus far, only 15 cases have been reported in English literature. Variceal bleeding from HJ is almost always due to pre-hepatic portal hypertension secondary to portal vein thrombosis or portal vein stenosis from local adhesions from previous surgery. Furthermore, repeated inflammation, such as recurrent retrograde cholangitis, bile leakage, or tumor compression or invasion, has also been implicated.[4,5] However, the reason why varices often form at the site of adhesions remains unclear. Ectopic varices at anastomosis may result in massive and

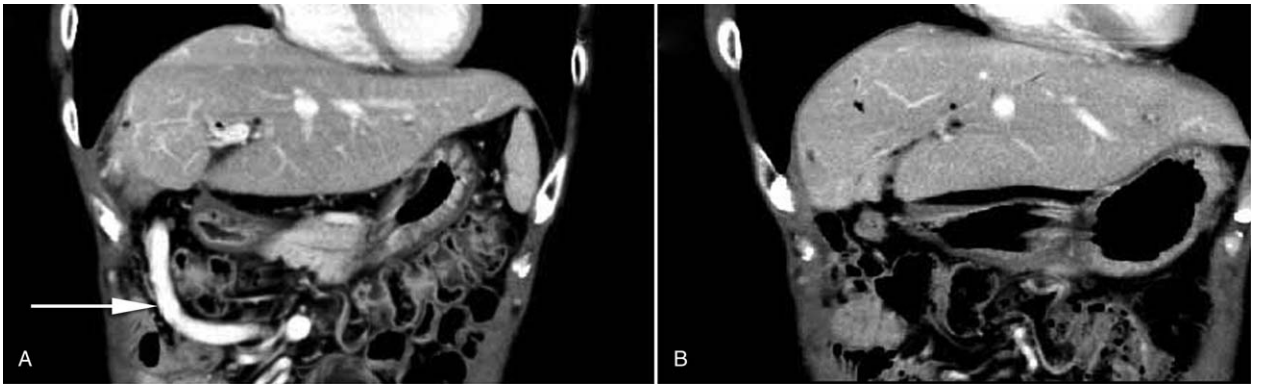

Figure 2. The blood vessel of the jejunal loop disappeared after operation. (A) Preoperative MDCT. White arrow, blood vessel of the jejunal loop, (B) postoperative MDCT. MDCT = multidetector computed tomography.

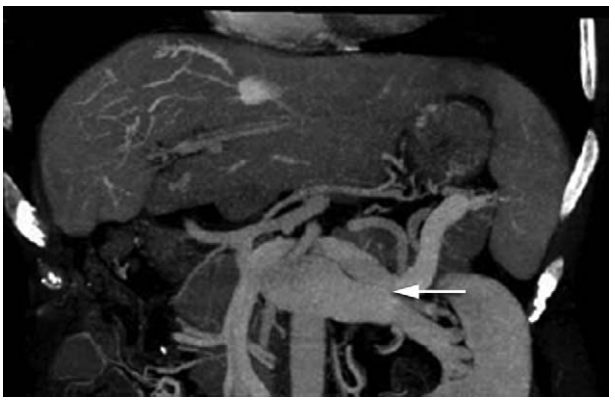

Figure 3. The anastomotic stoma of the splenic and left renal veins was visible and the distal part of the left renal vein evidently expansive. White arrow, anastomotic stoma of the splenic and left renal veins.

life-threatening bleeding.[4,5] Patients are usually asymptomatic except for repeated episodes of unexplained GI bleeding. The first episode may occur years after the initial operation.

In this report, the patient presented with early formation of portal vein thrombosis after the initial operation following the repair of the portal vein, which led to the narrowing of its lumen, leading to portal thrombosis. As a result of the 2 portal thrombectomies, the portal vein was finally occluded, which led to the development of cavernous transformation involving the whole length of the extrahepatic portal vein. Collaterals then developed at the anastomosis, with the intrahepatic portal vein system communicating with the mesenteric vein of the jejunal afferent loop, which eventually resulted in the formation of varices at the HJ anastomosis.

B ultrasound may shows occlusion of the portal vein and development of varices in some patients. Although computed tomography has only 72% diagnostic efficiency,[6–8] MDCT with portography is more valuable in diagnosis because it provides excellent images of portosystemic collaterals in patients with portal hypertension.[9–11] Technetium-99m-labeled red cell scintigraphy has no value in detecting the source of ectopic varices bleeding either. Arteriography is another frequently used modality. Since positive identification of the bleeding source necessitated a bleeding rate of at least 1 mL/min, it cannot show the source of the bleeding when the hemorrhage subsides.[12–14] When vasodilators, anticoagulants, or thrombolytic agents are given to induce hemorrhage, provocative angiography may identify 75% of the bleeding source.[13]

In this case, B ultrasound revealed the portal vein occlusion, cavernous transformation, and the development of collaterals and varices at the HJ anastomosis, distended splenic vein, and splenomegaly. MDCT portal venography confirmed these findings and provided clear images of the varices and multiple collaterals between the portal and mesenteric veins together with a significant dilation of the mesenteric vein of the jejunal afferent loop. Technetium-99m-labeled red cell scintigraphy and arteriography failed to identify the source of bleeding.

Although there is no standard management of jejunal varices that form at HJ anastomosis, the available treatment options are minimally invasive and surgical obliteration of varices including embolization, ligation, reanastomosis, and endoscopic therapy and decompressing operation including surgical shunt and transjugular intrahepatic portosystemic shunt (TIPS). Endoscopic therapy with sclerosant injection has been reported to be effective in controlling ectopic varices.[16] However, standard endoscopy will not reach the HJ anastomosis via Roux-en-Y. Of 15 reported cases, 3 patients underwent endoscopic sclerosant therapy as the initial treatment, but rebleeding of the varices occurred in 2 cases after the treatment 1 week and 1 month, respectively.[15–24] Secondary embolization of the dilated jejunal branches and surgical shunt were performed, which had successfully controlled the bleeding. Surgical ligation for bleeding anastomotic varices was reported in 1 case. Reanastomosis were reported in 2 cases by Ishida et al and Taniguchi et al in 1998 and 2008, with hepatic failure occurring in the last case.[4,25]

Surgical shunt is a radical treatment for controlling bleeding through decompression of the portal vein system. Chen et al[26] reported a proximal splenic–left intrahepatic portal vein shunt for extrahepatic portal vein obstruction. On angiography, disappearance of the collaterals was noted after the operation. The similar phenomenon was observed in our case, and there was no recurrence of bleeding or other complications after long term follow-up. Massive invasion, postoperative encephalopathy, and shunt occlusion were the main concerns of those who question this procedure.

To the best of our knowledge, this patient is the first reported case of unusual bleeding from hepaticojejunostomy controlled by a side-to-side splenorenal shunt. Ectopic variceal bleeding from HJ is unusual and difficult to manage. This is a useful treatment of a side-to-side venovenal anastomosis was created between splenic vein and left renal vein to form a shunt for decompression of varices at HJ. Surgical shunt remains effective treatment option for whom conservative management has failed to arrest bleeding which offers the most effective efficacy in reducing portal vein pressure and controlling varices bleeding from HJ.

Author contributions

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Writing – original draft: Meisheng Li, Zuojun Zhen.
Writing – review and editing: Huanwei Chen.

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