Clinical Case Report

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ERCP-related complication is not the only cause of GI bleeding in post-liver transplantation patients

A case report

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Abstract

Rationale: Endoscopic retrograde cholangiopancreatography (ERCP) is the treatment of choice for biliary complications in liver transplantation (LT) recipients as it is both diagnostic and therapeutic. The specific risks following ERCP among LT recipients have not been well studied.

Patient concerns: A 56-year-old man with a history of orthotopic LT underwent endoscopic retrograde cholangiopancreatography (ERCP) as a treatment of biliary strictures, whereby a plastic stent was implanted. Thirteen days after ERCP the patient developed multiple episodes of hematemesis.

Diagnosis: Digital subtraction angiography (DSA) of the hepatic artery and superior mesenteric artery showed a hepatic pseudoaneurysm (PA) in the left hepatic artery. The final diagnosis was bleeding from the PA.

Intervention: Interventional embolization of the branch with PA was performed to stop the bleeding.

Outcome: The patient remained free of GI bleeding for 25 days after interventional embolization, but he developed another bout of bleeding and unfortunately passed away.

Lessons: ERCP-related complication is not the only cause of post-ERCP bleeding, and that other primary causes should also be ruled out.

Abbreviations: DSA = digital subtraction angiography, ERCP = endoscopic retrograde cholangiopancreatography, EST = endoscopic sphincterotomy, GI = gastrointestinal, LT = liver transplantation, MRCP = magnetic resonance cholangiopancreatography, PA = pseudoaneurysm.

Keywords: ERCP, gastrointestinal bleeding, liver transplantation

1. Introduction

Biliary complications following orthotopic liver transplantation (LT), in which biliary strictures and leaks are the most common, are important causes of graft dysfunction and re-transplantation. Endoscopic retrograde cholangiopancreatography (ERCP) is the treatment of choice for biliary complications as it is both diagnostic and therapeutic.[1,2] The reported complication rate following ERCP among the general population varies between 4% and 12%.[3] These complications include acute pancreatitis, cholangitis, perforation, and bleeding due to sphincterotomy. Among these, acute pancreatitis is the most common, whereas gastrointestinal (GI) bleeding has rarely been reported.[4–6] The specific risks following ERCP among LT recipients have not been well studied. We herein report an unusual case of GI bleeding in a patient who underwent both LT and ERCP, and reviewed the procedure-related complications of ERCP post-LT (Table 1).[7–12]

2. Case presentation

A 56-year-old male patient with underlying severe chronic hepatitis B infection and liver failure underwent orthotopic LT, during which the hepatic arteries and portal veins were reconstructed and bile duct was duct-to-duct anastomosed. A bedside ultrasound on day 1 post-LT showed no abnormalities in the hepatic artery, portal vein, hepatic vein, or bile duct.

However, the patient's bilirubin level gradually increased from day 5 post-LT onwards. A repeat bedside ultrasound examination revealed dilatation of the intrahepatic bile duct. Magnetic resonance cholangio-pancreatography (MRCP) performed suggested the presence of dilated hepatic and intrahepatic bile ducts. Following an assessment by experienced endoscopists, we performed an ERCP on day 10 post-LT. A stricture was found on the upper segment of the common bile duct; we proceeded with an endoscopic sphincterotomy (EST), and a plastic "Christmas tree"-like biliary stent was placed (Fig. 1). The patient's serum biochemistry gradually improved, and the total

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Table 1

Procedure-related complications of ERCP postliver transplantation.

Authors	Year	Complications					
		Cholangitis	Pancreatitis	Bleeding	Bile leakage	Stent associated	Others
Seo et al[8]	2009	6.8%	10.3%	0%	3.4%	Not mention	Not mention
JChang et al[9]	2010	Immediate post-ERCP cholangitis 10.6% Delayed post-ERCP cholangitis or liver abscess 29.2%	8.2%	Not mention	Not mention	Not mention	Not mention
Balderramo et al[10]	2012	2.6%	5.3%	0.7%	Not mention	Not mention	Not mention
Hsieh et al[11]	2013	9.1%	1.6%	0.5%	Not mention	(Stent occlusion) 6.4%	liver abscess (1/187) duodenal perforation (1/187)
Chan et al[12]	2013	0%	0.8%	0.8%	Not mention	(Stent migration)1.6%	Not mention

ERCP = endoscopic retrograde cholangiopancreatography.

bilirubin level decreased post-ERCP (the total bilirubin and direct bilirubin levels normalized from 265 μmol/L and 178.6 μmol/L at their highest, respectively).

However, on day 13 post-ERCP, the patient developed multiple episodes of hematemesis, although his vital signs remained stable. A continuous infusion of PPI, somatostatin, and vasopressin was initiated, as well as blood transfusion. On day 15 post-ERCP, an upper GI endoscopy revealed multiple ulcers in the gastric fundus, along with mild esophageal varices, indicating that stress ulcers might have been the cause of the GI bleeding (Fig. 2). The duodenal mucosa appeared normal.

Despite treatment, the patient developed another episode of hematemesis on day 21 post-ERCP, with a sharp decline of hemoglobin from 99 g/L to 78 g/L. At this juncture, we postulated that the bleeding might have been due to ERCP. An emergency upper GI endoscopy revealed copious amount of blood clots surrounding the stent, along with oozing of blood at the duodenal papilla (Fig. 3). Considering that he was a post-LT patient, digital subtraction angiography (DSA) of the hepatic artery and superior mesenteric artery was conducted; a hepatic pseudoaneurysm (PA) measuring 11.6 mm × 9.7 mm was detected in the left hepatic artery (Fig. 4). Interventional embolization of the branch with PA

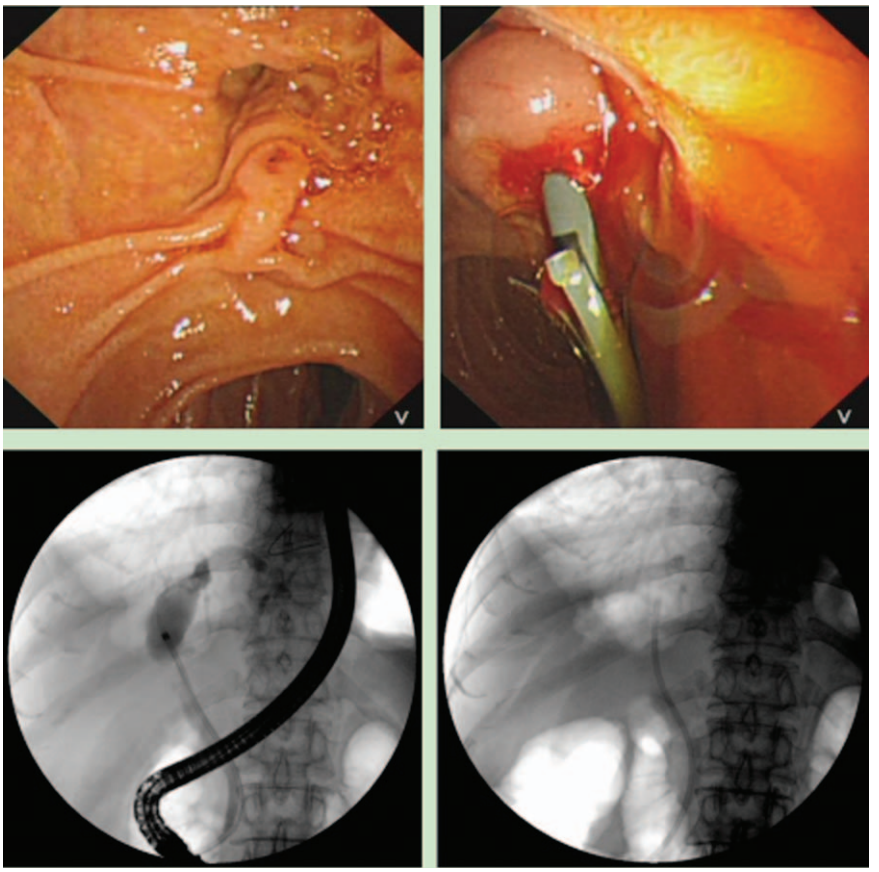

Figure 1. ERCP revealed a stricture in the upper segment of the common bile duct; an endoscopic sphincterotomy was performed, and a plastic stent was placed. ERCP = endoscopic retrograde cholangiopancreatography.

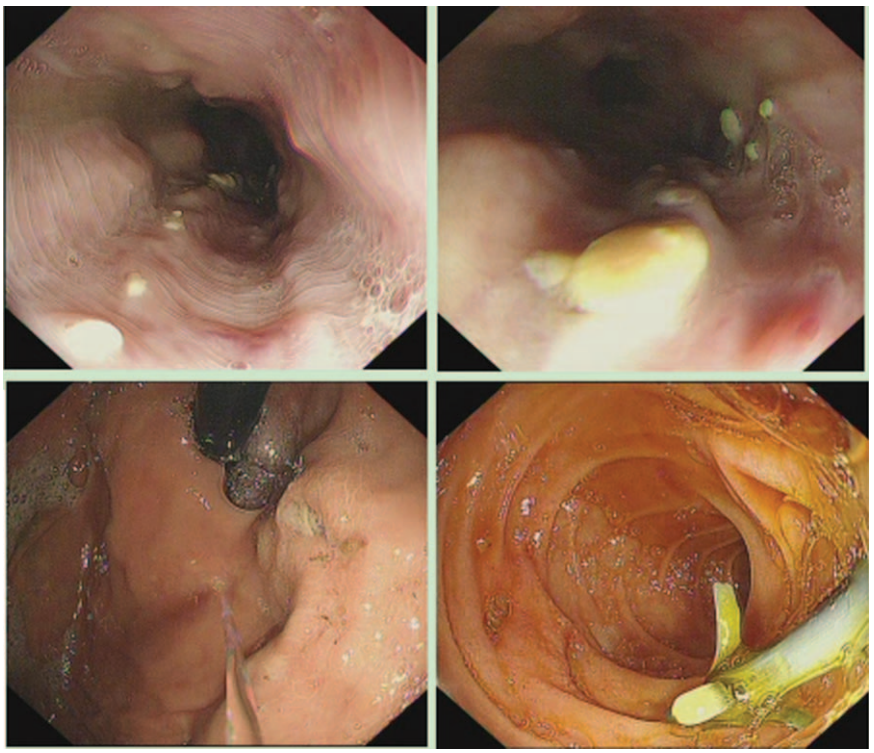

Figure 2. Upper GI endoscopy on day 15 post-ERCP revealed multiple ulcers in the gastric fundus, along with mild esophageal varices, but normal appearance of the duodenal mucosa. ERCP = endoscopic retrograde cholangiopancreatography, GI = gastrointestinal.

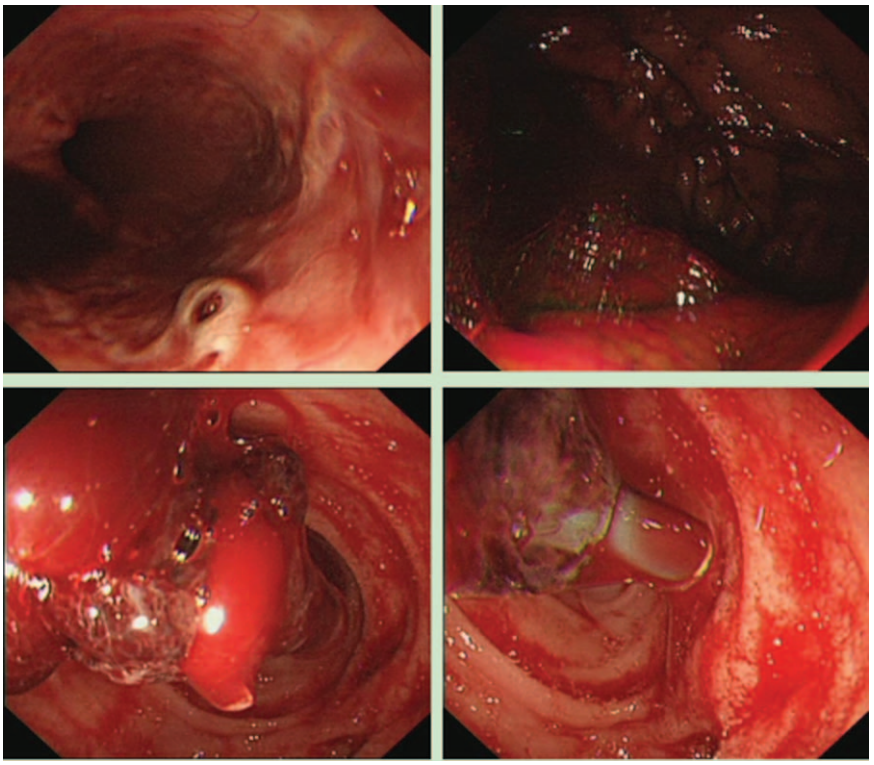

Figure 3. Upper GI endoscopy on day 21 post-ERCP revealed copious amount of blood clots surrounding the stent, with oozing of blood at the duodenal papilla. ERCP = endoscopic retrograde cholangiopancreatography, GI = gastrointestinal.

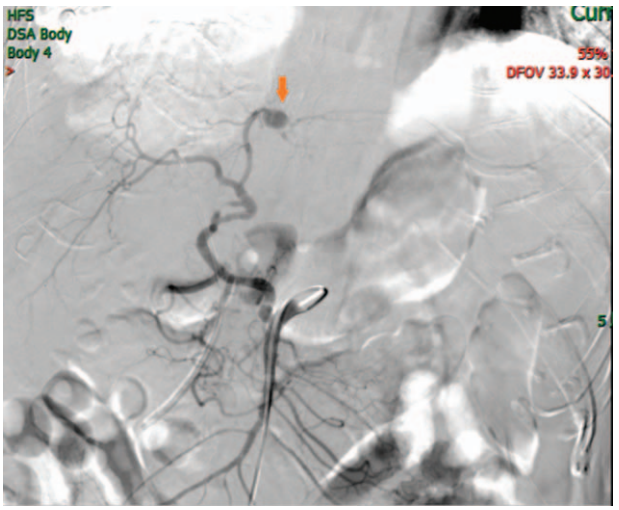

Figure 4. DSA detected rupture of a pseudoaneurysm of left hepatic artery (red arrow), with extravasation of contrast medium outlining. DSA = digital subtraction angiography.

was performed, and the patient remained free of GI bleeding for 25 days. Following this, he developed another bout of hematemesis, this time with melena. Another hepatic PA was discovered by DSA and interventional embolization was conducted for the second time. Two days later (day 57 post-LT), the patient developed bradycardia and hypotension, and spontaneous respiration stopped. Although there was some mild improvement in his vital signs following initiation of resuscitation efforts, the patient's family decided to forgo further aggressive resuscitation, and he subsequently passed away.

3. Discussion

ERCP is associated with several risks, including cholangitis, pancreatitis, bleeding, perforation, and, in rare cases, death.[4–6,13,14] Rare complications such as subcapsular hepatic hematoma have been reported in some cases.[15] A retrospective review of 11,497 ERCP procedures revealed that among 40 cases of bleeding, only 9 were severe.[4] In a meta-analysis of 21 prospective studies, the rate of post-ERCP bleeding was 1.3%, most of which were graded moderate.[3] Sphincterotomy is considered to be a high-risk procedure for post-ERCP bleeding, even though the rate of severe hemorrhage caused by sphincterotomy was less than 0.1%.[3] This suggests that severe hemorrhage post-ERCP is not very common. Unfortunately, there is lack of data regarding the occurrence time of bleeding post-ERCP, probably due to its low occurrence rate. Based on available data, post-ERCP bleeding may be immediate or delayed. In our experience, bleeding usually occurs intra-operatively or within 24 hours post-ERCP. In the present case, the patient's coagulation profile was normal following LT. With platelet transfusion, his mean platelet level was approximately 70 G/L (lowest: 47 G/L, highest: 122 G/L) during the period between ERCP and his first bleeding post-ERCP. Low platelet levels and EST may be the noteworthy risk factors for ERCP-associated bleeding in the present case.

Hepatic artery pseudoaneurysm (PA) post-LT is a potential life-threatening complication, with an incidence of approximately 2%.[1] The most common clinical manifestation of hepatic artery pseudoaneurysm is rupture with bleeding, such as GIbleeding or hemobilia or intra-abdominal hemorrhage. Local sepsis, biloma, and biliary tract infection may increase the risk of bleeding. Since it is usually difficult to treat a ruptured PA, early detection of it following LT is of great significance. Routine follow-up computed tomography (CT) or magnetic resonance imaging may help to detect PAs before they rupture, although angiography remains the gold standard for diagnosis. In our case, the PA was located in an extrahepatic artery, and the main manifestation was GI bleeding. The use of tacrolimus and biliary obstruction may contribute to the development of the PA. A CT angiography of the hepatic artery following LT (but before ERCP) showed presence of a small PA (measuring 1.9 mm $\times$ 3.9 mm) near the liver hilum; unfortunately, we did not pay close attention to this finding then. In addition, when the patient developed hemorrhage post-ERCP, we did not consider the primary disease and possible complications of LT.

Biliary tract-related complications following LT can be successfully managed with ERCP; however, the incidence and risk factors for post-ERCP complications among patients who have undergone LT have not been well studied. Balderramo et al[1] reported that the incidence of post-ERCP complications among LT patients is similar to that among the general population. Another study which compared post-ERCP complications between 1634 LT patients and 5852 concluded that post-ERCP bleeding was more frequent in the LT patients than the nontransplanted patients ($P < .05$).[16] Many factors, such as biliary sphincterotomy, renal failure, repeated pancreatic injections, and immunosuppressive therapy, may increase the risk of post-ERCP complications among LT patients.

To summarize, this was an unusual case of severe GI bleeding in an LT patient who received ERCP. We had initially misdiagnosed it as ERCP-associated bleeding; a second scope demonstrated the source of bleeding to be from a hepatic PA. We would like to emphasize the rare occurrence of ERCP-associated GI bleeding; other primary causes should therefore be ruled out, especially in delayed or severe cases.

4. Informed consent

The Ethics Committee of Tongji Medical College, Huazhong University of Science and Technology gave approval for this case report.

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