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CASE REPORT

Intrahepatic artery pseudoaneurysm associated with a metallic biliary stent after living donor liver transplantation: report of a case

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Abstract An intrahepatic artery pseudoaneurysm (IHAA) is a very rare but potentially lethal complication occurring after liver transplantation. This report presents a case of an IHAA associated with a metallic biliary stent after liver transplantation. A 40-year-old male underwent living donor liver transplantation (LDLT) using a left lobe graft. The bile duct reconstruction was performed with Roux-en-Y hepaticojejunostomy. He developed obstructive jaundice 5 years after LDLT, and had biliary stricture of the anastomosis area, therefore, the two metallic biliary stents were finally positioned at the stricture of the biliary tract. He suddenly developed hematemesis 8 years after LDLT, and computerized tomography scan showed an IHAA. Although seven interlocking detachable coils were placed at the neck of the aneurysm, hematemesis recurred 3 days after the initial embolization. Therefore, retransplantation was successfully performed 25 days after the embolization of IHAA using a right lobe graft from his son. In conclusion, metal stent insertion can lead to the fatal complication of HAA. The placement of a metallic stent could have been avoided in this case. Percutaneous metallic stent insertion for biliary stenosis after liver transplantation should therefore only be performed in carefully selected patients.

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Abbreviations

HAA	Hepatic artery pseudoaneurysm
IHAA	Intrahepatic artery pseudoaneurysm
LDLT	Living donor liver transplantation
POD	Postoperative day
CT	Computerized tomography
AST	Aspartate aminotransferase
ALT	Alanine aminotransferase
HCV	Hepatitis C virus
IDC	Interlocking detachable coil
PT-INR	Prothrombin time-international normalized ratio

Introduction

An intrahepatic artery pseudoaneurysm (IHAA) is a very rare occurrence and can present with nonspecific abdominal pain or massive gastrointestinal bleeding due to hemobilia or perforation. A series of 74 patients with liver transplantation that experienced gastrointestinal bleeding, included two cases that were secondary to hemobilia: one was secondary to a liver biopsy and the other to a hepatic artery pseudoaneurysm (HAA) [1]. The use of interventional procedures is also associated with an increased incidence of hemobilia. HAA accounts for nearly 10 % of hemobilia cases [2]. Hemobilia and HAA are associated with percutaneously placed stents and endoscopically placed metal stents [3, 4]. This report presents a case of an IHAA associated with a metallic biliary stent after liver transplantation.

Case report

A 40-year-old male underwent living donor liver transplantation (LDLT) for end-stage liver disease due to hepatitis C in 2000. The donor was his 38-year-old brother, who donated his left hepatic lobe. The bile duct reconstruction was performed with Roux-en-Y hepaticojejunostomy. Portal vein thrombosis was diagnosed on post-operative day (POD) 1, and thrombectomy of the portal vein was successfully performed. Significant collateral shunt vessels were ligated to increase the portal vein flow. Immunosuppression was administered with tacrolimus and prednisone. He recovered from the procedure and was discharged at POD 55. He developed obstructive jaundice and moderate weight loss 5 years after LDLT. Computerized tomography (CT) showed intrahepatic bile duct dilatation. He had biliary stricture of the anastomosis area. Therefore, the two metallic biliary stents were percutaneously positioned at the stricture of the biliary tract. A liver biopsy taken at that time revealed recurrent hepatitis C with severe fibrosis. Therefore, a combination therapy of peg-interferon (Peg-Intron®, Schering-Plough Corporation, NJ, USA) and ribavirin (Rebetol®, Schering-Plough Corporation, NJ, USA) was started, and end-of-treatment response (EOTR) was obtained after 48 weeks therapy. However, liver function tests such as aspartate aminotransferase (AST) and alanine aminotransferase (ALT) value elevated and HCV-RNA from serum was positive again 3 months after the cessation of interferon therapy. The combination therapy of peg-interferon and ribavirin was resumed and continued until recently. He suddenly developed hematemesis 8 years after LDLT, and laboratory data showed that his total bilirubin was 16.1 mg/dL, hemoglobin 7.9 g/dL, hematocrit 23.3 %, albumin 2.6 g/dL, and prothrombin time 13.4 s. There were no signs of either bacterial or mycotic infections. CT showed a 38 × 24 mm IHAA as well as portal vein thrombosis (Fig. 1a, b). Emergent abdominal angiography confirmed a pseudoaneurysm at the proximal portion of the segment 3 branch of the left hepatic artery, where the tip of the two previous metallic biliary stents was positioned. Seven interlocking detachable coils (IDC; Boston Scientific Corporation, MA, one IDC soft; 2 × 4 mm, three IDC soft; 4 × 80 mm coils, two IDC soft; 4 × 120 mm, one FPC18; diamond 2-3-2 mm) were placed at the neck of the aneurysm and in the HAA (Fig. 2). Laboratory data before coil embolization showed total bilirubin 17.4 mg/dL, AST 74 U/L, ALT 47 U/L, hemoglobin 10.4 g/dL, hematocrit 24.5 %, albumin 2.4 g/dL, and prothrombin time 14.1 s (INR 1.24). Laboratory data 1 day after the procedure showed total bilirubin 16.0 mg/dL, AST 88 U/L, ALT 53 U/L, hemoglobin 8.8 g/dL, hematocrit 24.5 %, albumin 2.1 g/dL, and prothrombin time 14.3 s (INR 1.26). The

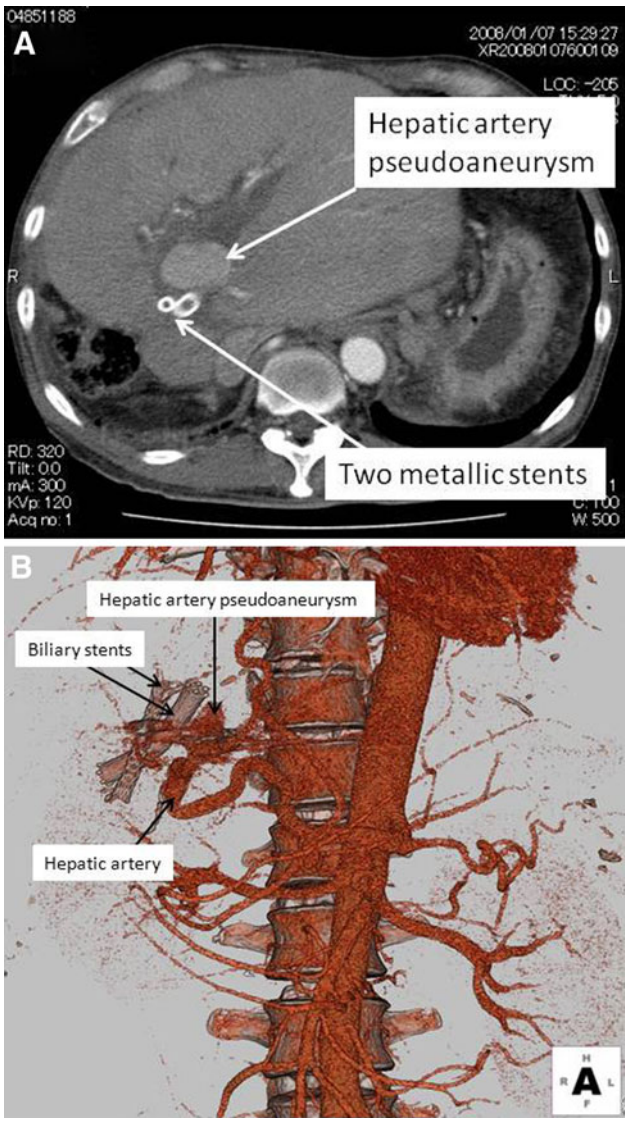

Fig. 1 **a** Intrahepatic artery pseudoaneurysm seen in a contrast-enhanced computed tomography scan. The hepatic artery pseudoaneurysm was located near the two metallic stents in the graft liver. **b** Three-dimensional reconstructed CT images demonstrated the intrahepatic pseudoaneurysm in the hepatic artery. Two biliary stents were also noted close to the intrahepatic pseudoaneurysm

embolization of the HAA had not improved the liver function. However, hematemesis through the biliary tract recurred 3 days after the initial embolization. CT revealed another 20 × 18 mm IHAA at the distal portion of the segment 3 branch of the left hepatic artery near the coiled IHAA. Therefore, retransplantation was successfully performed 25 days after the embolization of IHAA, using a right lobe graft from his son. The findings of the one cutting section of the explanted graft liver are shown in Fig. 3. The HAA was located near the cut orifice of the segment 3 branch of the left hepatic artery, as shown in Fig. 1. Two metallic stents were at the orifice of hepaticojejunostomy.

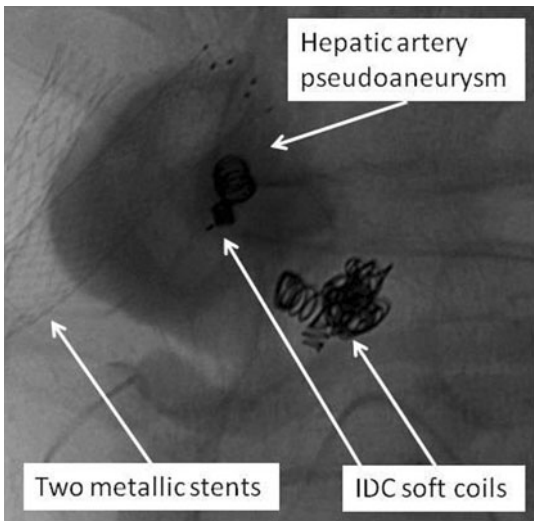

Fig. 2 Hepatic artery angiography after angioplasty and coil embolization. Seven IDC coils were placed at the neck of the aneurysm and in the hepatic artery pseudoaneurysm

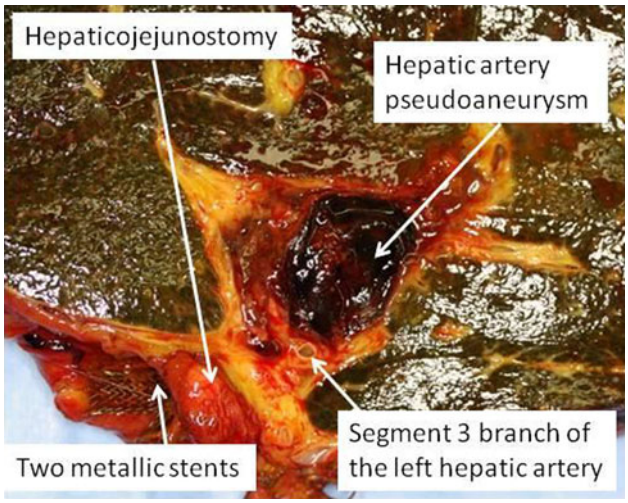

Fig. 3 A cut section of the explanted graft liver. The hepatic artery pseudoaneurysm was located near the segment 3 branch of the left hepatic artery. Two metallic stents were shown at the orifice of hepaticojejunostomy

A pathological examination of the removed graft liver revealed stage F3 fibrosis, dissection of the hepatic artery wall between the intimal layer and the internal elastic lamina with fibrous deposits around the HAA, and cholangitis with lymphocytic invasion and fibrotic changes in the wall of the hepatic duct.

Discussion

A HAA occurs in 1–2 % of liver transplantation patients [5, 6]. One of the major causes is surgical intervention to and around the hepatic artery. Another cause is the bacterial or mycotic infection. HAAs are classified as intrahepatic or extrahepatic depending on their location, with 20 % being intrahepatic and 80 % extrahepatic [3]. Intrahepatic pseudoaneurysms are usually related to previous procedures such as liver biopsy or percutaneous transhepatic biliary drainage. Extrahepatic pseudoaneurysms have a different pathogenesis, and local sepsis is the most important risk factor [3]. The present case of intrahepatic pseudoaneurysm may have had a ruptured bile duct. The inflammation surrounding the bile duct and adhesions between the metallic stent and the hepatic artery may have contributed to the formation of the pseudoaneurysm. There had been previous episodes of melena, which probably indicated bleeding from the pseudoaneurysm. There were no significant pathological findings or any evidence to indicate the cause of the HAA. The pathological examination showed cholangitis, but there were no clinical signs of cholangitis such as high-grade fever before retransplantation.

The stent was probably the cause of the aneurysm in this case, because the tip of the stent was at the site of aneurysm. There were no clinical signs of either bacterial or mycotic infections. Liver biopsies for HCV recurrence may have caused HAA, but the patient did not undergo liver biopsies after metallic stenting. Percutaneous treatment with metallic stents has a high risk for obstruction after a short to mid-term follow-up; however, the patient had recurrent hepatitis C and demonstrated liver cirrhosis. The indication for metallic stent insertion under potentially benign conditions was thought to be controversial, but he finally agreed and chose the option of the metallic stents after a percutaneous dilatation of the stenosis with internal–external stenting based on the patient's general condition.

The episode of hematemesis was associated with undetectable portal vein flow, suggesting increased compensatory hepatic artery flow, and this could lead to the formation of an aneurysm. A decreased portal venous flow causes increased hepatic arterial flow under experimental conditions [7, 8]. Direct or partial occlusion causes a significant decrease in portal vein flow without causing a decrease in systemic blood pressure, and increases the flow in the hepatic artery [7]. The growth and rupture of an intrahepatic aneurysm associated with hypertension, which caused increased hepatic artery flow, have also been reported [9]. Therefore, the metallic stent probably induced the aneurysm of the hepatic artery in combination with increased hepatic artery flow following the disappearance of the portal vein flow. The portal vein thrombosis may have been caused by liver cirrhosis and portal hypertension but not metallic stent insertion because the pathological examination showed that the metallic stent had no direct effect on the portal vein.

Bleeding from the aneurysm through the biliary tract around started 3 years after the metallic stents were positioned. Treatment options focus on catheter-based minimally invasive endovascular treatments such as coil embolization or covered stent prosthesis, especially in the event of rupture and hemobilia [10, 11]. Other options include retransplantation or ligation of the hepatic artery [4, 5]. These standard treatments are associated with a 69 % mortality rate [5, 10, 11]. The current aneurysm occurred within the hepatic graft and could not be surgically approached. In addition, the demonstrated liver cirrhosis was due to recurrent hepatitis C and portal vein thrombosis. The IHAA was embolized to obtain transient hemostasis, and retransplant was successfully performed 25 days after the procedure. Care should therefore be taken with the technique used for percutaneous biliary drain placement in order to prevent HAA formation. Plans for transhepatic needle access to the biliary tract should avoid the central bile ducts as much as possible. Central bile ducts are accompanied by larger and more crowded blood vessels, and the likelihood of transgressing an artery or a large vein is therefore higher in this area [12].

In conclusion, metal stent insertion can lead to the fatal complication of an HAA. Placement of a metallic stent could have been avoided in the current case. Percutaneous metallic stent insertion for biliary stenosis after liver transplantation should therefore only be performed in carefully selected patients.

Conflict of interest All authors have no conflict of interest to declare.

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