



# Management of a Portobiliary Fistula and Obstructive Biliopathy in a Split Liver Graft Transplant Recipient Posthepaticojejunostomy with Portal Stent Graft

Saurabh Kumar<sup>1</sup> Apoorva Batra<sup>1</sup> Vaibhaw Kumar<sup>2</sup> Vivek Vij<sup>2</sup>

<sup>1</sup>Department of Interventional Radiology, Fortis Memorial Research Institute, Gurugram, Haryana, India

<sup>2</sup>Department of Liver Transplant and Hepatobiliary Surgery, Fortis Memorial Research Institute, Gurugram, Haryana, India

**Address for correspondence** Dr. Saurabh Kumar, MD, DNB, FGIR, Department of Interventional Radiology, Fortis Memorial Research Institute, Sector 44, Gurugram 122002, Haryana, India (e-mail: sunny\_mamc911@yahoo.com).

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## Abstract

### Keywords

- ▶ portobiliary fistula
- ▶ liver transplant
- ▶ hepaticojejunostomy

Biliary complications following liver transplantations, in the form of bile leak and bile duct strictures, are well recognized. Portobiliary fistulas in the transplant setting are rare, but may cause potentially life-threatening complications. The presented case shows the successful treatment of a hilar portobiliary fistula in a transplanted liver posthepaticojejunostomy with portal vein stent graft followed by biliary drainage with percutaneous hepaticojejunostomy stricture dilatation.

## Case Report

A 60-year-old man with nonalcoholic steatohepatitis (NASH) related cirrhosis underwent right split graft liver transplantation followed by endoscopic biliary balloon dilation and placement of plastic stents multiple times, over a period of 6 months for biliary anastomotic stricture. Hepaticojejunostomy (HJ) was offered due to persistence of obstructive biliopathy symptoms and was successfully performed after hepatic duct mobilization. Two weeks later, the patient got admitted in the emergency department with symptoms of fever and hypotension with one episode of melena. Blood count showed hemoglobin (Hb) 9 g/dL, total leucocyte count (TLC) of 16,000/mm<sup>3</sup>, and deranged liver function (serum bilirubin: 5 mg/dL; serum alkaline phosphatase [ALP]: 400 IU/L; serum gamma-glutamyl transferase [GGT]: 350 IU/L; alanine aminotransferase [ALT]: 86 U/L; and aspartate transferase [AST]: 88 U/L). Ultrasound followed by contrast magnetic resonance imaging (MRI) showed dilated intrahepatic bile ducts (predominant posterior segment) with intrahepatic cystic biloma (▶ **Fig. 1**). Based on imaging findings, obstructive

biliopathy secondary to HJ site stricture and superadded cholangitis was considered and percutaneous biliary drainage was planned for the patient.

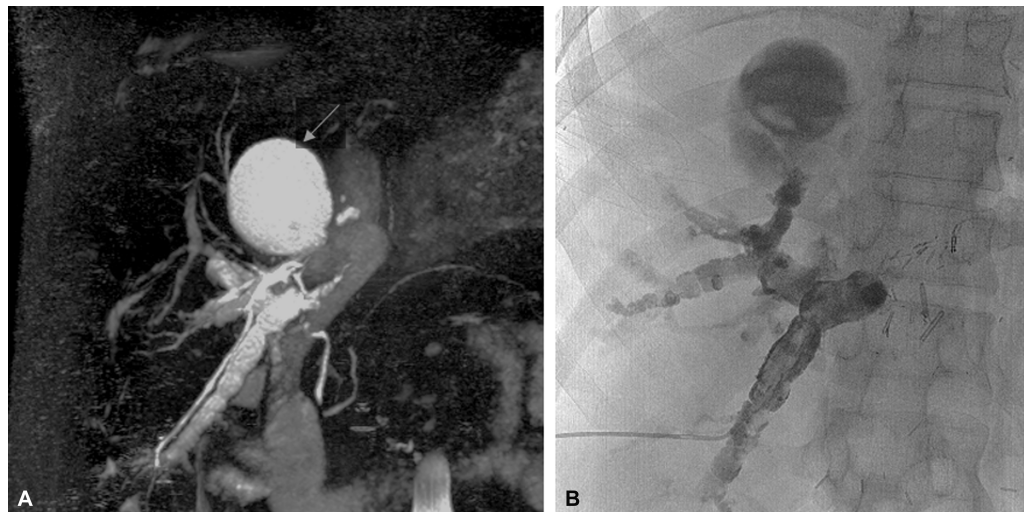
The posterior segment duct was punctured under ultrasound guidance with a 4-Fr micropuncture set (Cook Medical, Bloomington, United States). Then Terumo wire (Glidewire; Terumo Interventional Systems) was negotiated fluoroscopically across the duct followed by advancement of a 5-Fr Kumpe catheter (Cook Medical) over it. Cholangiogram through the catheter showed opacification of the portal vein (▶ **Fig. 2A**), thereby confirming the diagnosis of portobiliary fistula. In spite of multiple attempts, the wire could not be negotiated across the HJ stricture site into the jejunum as it kept going into the portal vein. Therefore, it was decided to first seal off the fistula site by placing a covered stent across the portal vein. The anterior segmental peripheral portal vein branch was then punctured and the portogram (▶ **Fig. 2B**) revealed normal anterior segmental branches with nonvisualization of the posterior segmental portal vein. Covered Fluency plus stent 12 mm × 60 mm (BD; Covington, United States) was placed in the main portal vein across the fistula site followed by coil

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**Fig. 1** (A) Coronal magnetic resonance cholangiopancreatography (MRCP) maximum intensity projection (MIP) image showing dilated intrahepatic biliary radicles predominantly of the posterior segment with the cystic structure representing biloma (*small arrow*). (B) Fluoroscopic cholangiogram image showing opacification of the dilated biliary ducts and biloma cavity.



**Fig. 2** (A) Digital subtraction angiographic image showing opacification of the main portal vein (*small arrow*) suggestive of portobiliary fistula. (B) Digital subtraction angiographic image after insertion of the covered stent showing normal portogram. (C) Fluoroscopic cholangiogram image after portal vein stenting showing nonopacification of the portal branches suggesting fistula closure.

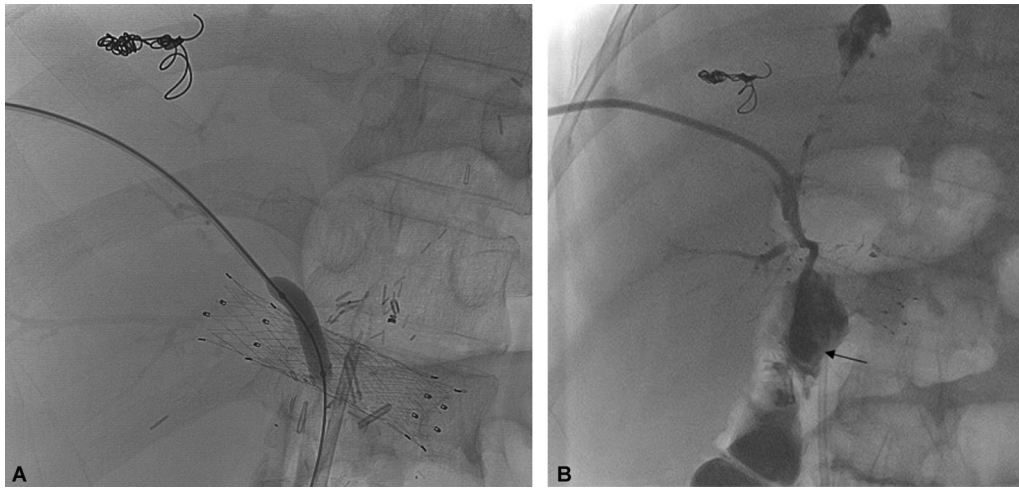
embolization (Nester pushable coils, 8 mm × 14 mm) of the portal vein entry liver tract. The final cholangiogram showed nonopacification of the portal vein suggesting closure of the fistula site (→ **Fig. 2C**). The patient was discharged with external biliary catheters and was called back after 2 weeks.

The patient was again taken up for HJ stricture negotiation. This time internalization was successful with balloon dilatation of HJ stricture was done with a 6 × 40 mm balloon (35LP Low Profile, Cook Medical) and placement of an 8-Fr internal external catheter (Ring Biliary catheter, Cook Medical) across (→ **Fig. 3**). Upgradation of the internal external catheter to 12 Fr was done after 2 weeks and the patient was discharged. On subsequent monthly follow-ups, the patient is doing fine with normal liver function and without any further episode of cholangitis.

HJ<sup>1</sup> is a viable option for a patient with endoscopic intervention failure besides percutaneous/cholangioscopic interventions and is the more effective method in the management of cases with complicated biliary complications, such as necrosis of the bile duct, biliary duct stones, and sludge especially in posttransplant settings. Portobiliary fistula<sup>2</sup> is a rare complication of liver transplantation, precipitating from

bile duct ischemia, infection, inflammation, or stone erosion. Other documented causes include malignancy, portal vein aneurysm, biliary stent insertion, liver biopsy, and biliary surgery. Clinically portobiliary fistula commonly presents with features of hemobilia. In our case, the likely precipitating factors for portobiliary fistula formation are previous multiple biliary interventions in the form of balloon dilatation and stenting, recurrent cholangitis episodes, and finally HJ surgery, which involves hepatic duct mobilization, with superadded infection in surgical bed leading to fistula formation and thrombotic occlusion of the posterior segmental portal vein.

Treatment options for portobiliary fistulas<sup>3</sup> include closure with surgery, coiling of the communicating portal vein branch, and balloon occlusion or covered stent placement within the associated portal vein or biliary branch. Since in our case the fistula site is close to hilum and we were not able to cross across HJ stricture, we decided to place the stent graft in the portal vein to seal the fistula site. Few isolated case reports of using portal stent grafts in posttransplant settings for portobiliary fistula have been described. Peynircioglu and Cwikel<sup>2</sup> described a portobiliary fistula secondary to hepatic arterial thrombosis



**Fig. 3** (A) Fluoroscopic image showing balloon dilatation (*small arrow*) of hepaticojejunostomy stricture site. (B) Fluoroscopic cholangiogram image taken through an internalized biliary catheter showing opacification of the jejunal loops (*small arrow*).

(HAT) after liver transplantation involving the peripheral bile duct and was treated by placing a Wallgraft in the portal vein. Stent graft placement is advantageous over the biliary approach secondary to high patency rates of portal stents. Placement of the covered biliary stent for portobiliary fistulas in liver transplant settings has been described by Murray et al,<sup>3</sup> which forms the other treatment option in such settings. After closure of the portobiliary fistula, the next step was to negotiate across HJ stricture with balloon dilatation and progressive placement of an upsized external internal drainage biliary catheter, which is the standard of care<sup>4</sup> of HJ stricture in liver transplant patients, thus obviating the need of re-surgery or re-transplantation.

This case highlights the importance of recognizing potential life-threatening complications of hilar portobiliary fistula in liver transplant recipients with HJ, which requires management in the form of fistula closure through placement of portal or biliary stent graft followed by percutaneous management of HJ site biliary stricture.

#### Funding

None.

#### Conflict of Interest

None declared.

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