Porto-Biliary Fistula Management with Endoscopy and Liver Transplantation

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Background	Portobiliary fistulas are a rare cause of hemobilia at times resulting from endoscopic procedures and their associated iatrogenic injuries. Additionally, accidental cannulation and/or visualization of the portal vein during endoscopic procedures are exceedingly rare, with a reported incidence of 1 in 6,000 to 8,000 cases.
Summary	We report a case of hemobilia resulting from a porto-biliary fistula secondary to iatrogenic complication of endoscopic retrograde cholangiopancreatography occurring in a cirrhotic patient, temporized with endoscopic stenting, and definitively treated at the time of liver transplantation.
Conclusion	To our knowledge, this is the first case report of the intraoperative localization and surgical management of a porto-biliary fistula during orthotopic liver transplantation.
Key Words	clinical practice; hemobilia; porto-biliary fistula; bilioportal fistula; liver transplantation

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The authors have no conflicts of interest to disclose.

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Case Description

Haemobilia is commonly defined as bleeding into the biliary system. It was first described in 1654 as a triad of upper abdominal pain, upper gastrointestinal hemorrhage, and jaundice.[1] Causes include portal hypertension of varying etiologies, trauma, and conditions in which the vascularity of the liver or pancreas may become disrupted into the biliary system. Still, iatrogenic etiologies account for most of the reported cases.[2,3] Porto-biliary fistula (PBF) is a rare cause of hemobilia, often resulting from endoscopic procedures and their associated iatrogenic injuries.[2,4] Reported methods of hemorrhagic stasis from PBF include endoscopic techniques of coagulation, hemoclipping, and stenting.[5] We report a case of hemobilia secondary to PBF in a cirrhotic patient, which was temporized with endoscopic stenting and definitively treated at the time of liver transplantation.

The patient is a 70-year-old male with cryptogenic cirrhosis initially diagnosed in 2008 during a routine laparoscopic cholecystectomy. In April 2017, a diagnosis of extra-hepatic portal vein thrombosis, biliary dilatation, and hepatocellular carcinoma was made. At this time, his thrombosed extra-hepatic portal vein was noted to be reconstituting distal to the coronary vein, with extra-hepatic portal vein patency at left and right portal veins. The biliary dilation caused by a portal biliopathy was managed with endoscopic retrograde cholangiopancreatography (ERCP) and biliary stenting. The HCC lesions were successfully treated with transarterial chemo-embolization (TACE).

The following two months were complicated by multiple episodes of cholangitis requiring repeat endoscopic interventions. In August 2017, the patient was admitted to the hospital for hemodynamic shock, cholangitis, and gastrointestinal bleeding, which were appropriately treated with antibiotics and blood transfusions. Unfortunately, hepatic decompensation was now noted as he now had a model for end-stage liver disease (MELD) score of 33 and was subsequently placed on the liver transplant waitlist.

During this admission, a second episode of severe gastrointestinal bleeding urgently managed with esophagogastroduodenoscopy demonstrated bleeding from the major duodenal ampulla. ERCP demonstrated the opacification of the main portal vein located 3 cm to 4 cm from the ampulla (Figure 1A and Figure 1B), confirming the diagnosis of a PBF. The bleeding was controlled with a 15 mm extraction balloon, followed by the placement of a 10 mm x 8 cm fully covered Viabil® metal stent into the common bile duct. Stenting across the fistula allowed for tamponade of the fistulous defect and temporarily controlled bleeding. The patient underwent orthotopic liver transplantation with a cadaveric liver four days later.

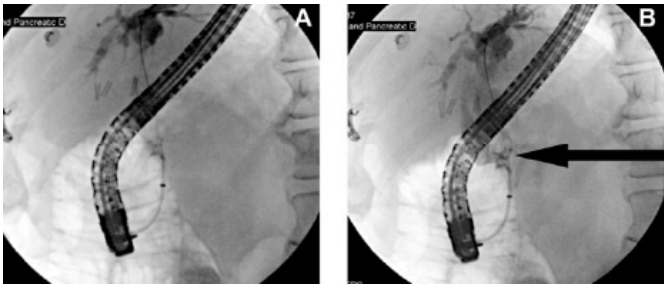

Figure 1. ERCP cholangiogram; arrow points to site of porto-biliary fistulous tract

Intraoperative findings during the liver transplantation revealed diffuse inflammatory changes at the hepatic hilum yielding the extra-hepatic portal vein and bile duct inseparable. The common bile duct was first incised, and the metallic stent visualized. Following manual stent extraction, rapid bleeding was temporarily controlled with digital compression of the portal vein at the level of the duodenum. A 1 cm porto-biliary fistula was identified on the posterior wall of the common bile duct (Figure 2).

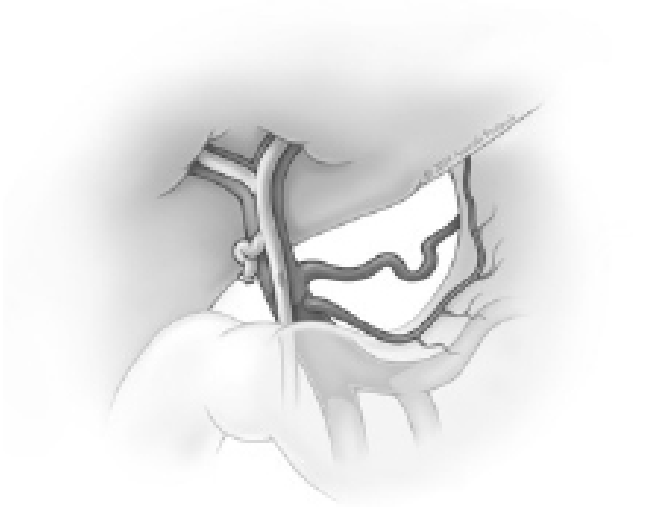

Figure 2. Porto-biliary fistula in situ. ©2019 Amanda Frataccia. Image used with permission

Division of the portal vein below the fistula was not a feasible option because this portion of the portal vein was necessary to provide the new liver with portal flow. The defect was then primarily repaired via the posterior wall of the common bile duct. The repair was completed with 4-0 Prolene suture in a running fashion while maintaining simultaneous manual compression (Figure 3 and Figure 4). Carefully avoiding narrowing of the portal vein during the repair, the common bile duct was transected distal and proximal to the site of the repaired fistula and oversewn with 4-0 Prolene suture. The anterior wall of the common bile duct was then excised, and the posterior wall left behind, attached to the native portal vein.

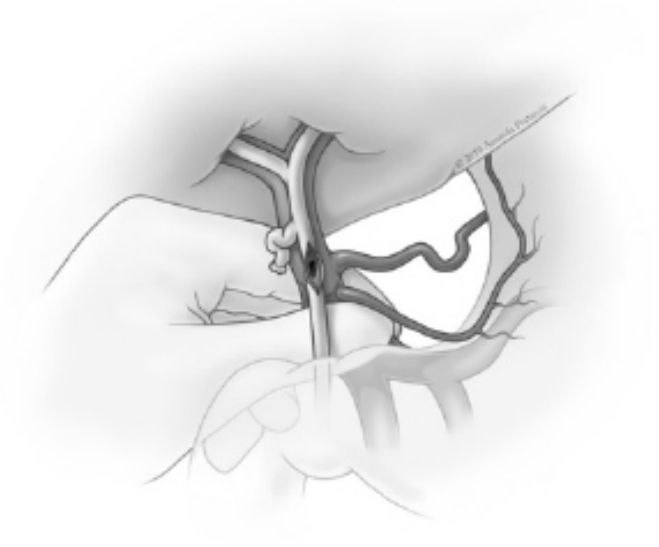

Figure 3. Porto-biliary fistula exposed prior to repair. ©2019 Amanda Frataccia. Image used with permission

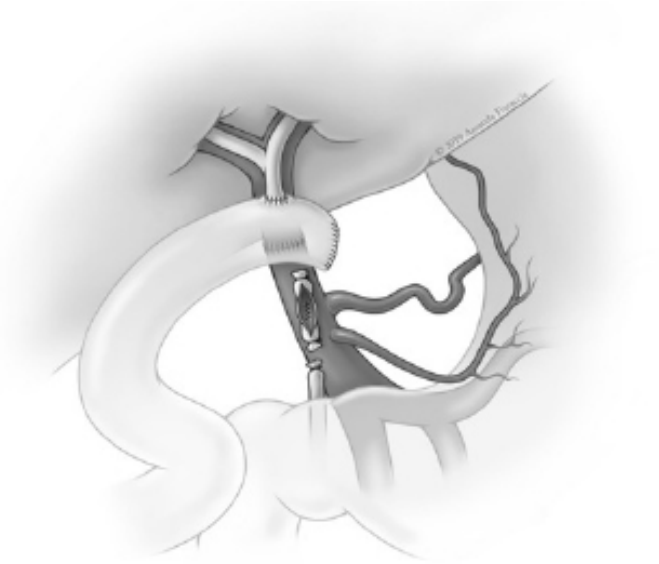

Figure 4. Porto-biliary fistula repair with simultaneous liver transplantation. ©2019 Amanda Frataccia. Image used with permission

The donor liver was implanted with donor and recipient portal veins anastomosed immediately above the site of the previously repaired PBF. Arterial inflow was established using an infrarenal aortic graft to the donor hepatic artery, and a hepaticojejunostomy was used for biliary drainage. The donor liver graft ischemia time was 428 minutes from the donor patient cross-clamp to graft arterial reperfusion in the recipient and 94 minutes from out-of-donor patient to graft arterial reperfusion in the recipient. The postoperative course was uncomplicated, and the patient was discharged to home on postoperative day 8. Two years after the transplant, the patient is alive and remains without major post-transplant complications.

Discussion

Porto-biliary fistulas are an uncommon cause of hemobilia. The etiologies are numerous but commonly include iatrogenic injury during percutaneous or endoscopic instrumentation.[3,6] Reported series describes injury during percutaneous hepatobiliary drainage, aberrant radiofrequency ablation, TIPS placement, and ERCP. Intraoperative surgical injuries from iatrogenic insults during choledochoduodenostomy, laparoscopic and open cholecystectomy, liver biopsy, and liver transplantation have also been reported.[6–9] The less common non-iatrogenic causes include choledocholithiasis, pancreatitis, liver abscesses, and hyper-vascular lesions invading the portal system.[7] Most PBF are amenable to radiological and endoscopic interventions. Still, surgical management may be indicated and include the primary repair of the fistula with portal vein resection and even the need for partial hepatectomy.

Conclusion

To the best of our knowledge, this is the first case report describing the intraoperative localization and primary repair of a previously stented porto-biliary fistula arising in the setting of portal biliopathy and cirrhosis.

Lessons Learned

Although performed during orthotopic liver transplantation, the technique described provides a viable approach to surgical repair in other instances of extra-hepatic porto-biliary fistula.

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