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CASE REPORT | BILIARY

Percutaneous Transhepatic Single-Operator Cholangioscopy-Guided Intraductal Stone Therapy in a Liver Transplant Patient With Ischemic Cholangiopathy

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ABSTRACT

Ischemic cholangiopathy is a feared complication after liver transplantation. We present a 68-year-old man who is status post-orthotopic liver transplant from a donation after cardiac death. His posttransplant course was complicated by the development of a biliary anastomotic stricture, ischemic cholangiopathy, biloma, recurrent cholangitis, and intrahepatic stones. Through the use of antegrade cholangioscopy with a single-operator cholangioscope (SpyGlass 2; Boston Scientific, Boston, MA) passed through a percutaneous sheath, we were able to visualize impacted stones within the left intrahepatic system and treat them using electro-hydraulic lithotripsy for stone fragmentation and removal.

INTRODUCTION

Advancements in endoscopic retrograde cholangiopancreatography (ERCP) have revolutionized the management of biliary stones. However, endoscopic management of intrahepatic stones continues to pose a challenge for clinicians. We discuss a patient with intrahepatic stones who was treated with percutaneous transhepatic cholangioscopy with electrohydraulic lithotripsy (EHL).

CASE REPORT

A 68-year-old man with alcoholic cirrhosis underwent an orthotopic liver transplant from a donation after cardiac death (DCD). The donor was 29 years old without any known underlying liver disease or steatosis. Cold ischemia time for the donor liver was 5 hours 42 minutes. From the time the liver was removed from ice to establishment of portal inflow was 45 minutes. Six months after liver transplant, the recipient was found to have developed a biliary anastomotic stricture and intrahepatic changes consistent with ischemic cholangiopathy. His graft was not complicated by hepatic artery thrombosis. In the setting of an anastomotic stricture and ischemic cholangiopathy, he developed cholangitis and a biloma. For management, he underwent ERCP with biliary stent placement and external biliary drain for management of his biloma. Over the next 2 years, he had recurrent episodes of cholangitis and bilomas requiring multiple drain placements, ERCPs with stent exchange and placement on an internal-external biliary drain (18Fr × 40 cm; Cook Medical , Bloomington, IN). An MRCP 36 months after his transplant demonstrated persistent biloma and mural irregularities within the intrahepatic ducts concerning for intrahepatic stones (Figures 1 and 2). Because of recurrent cholangitis and a failure to improve with traditional management, a combined interventional radiology (IR)-biliary endoscopic procedure was undertaken 36 months after his transplant.

IR removed the existing biliary drain over a 0.035" × 180 cm Amplatz Ultra-stiff guide wire (Cook Medical ; Bloomington). A 14 Fr × 45 cm Check-Flo sheath (Cook Medical ; Bloomington) was then advanced into right intrahepatic bile duct for stone extraction. A 5 Fr × 65 cm Berenstein catheter (AngioDynamics) was used to select left-sided bile ducts. Stones were removed by inflating a 5 Fr × 80 cm Fogarty embolectomy catheter (Edwards Lifesciences, Irvine, CA) in distal left bile ducts and pulling the stones into the common hepatic duct. Next, the Fogarty balloon was used to push the stones through the CBD past the ampulla. Antegrade

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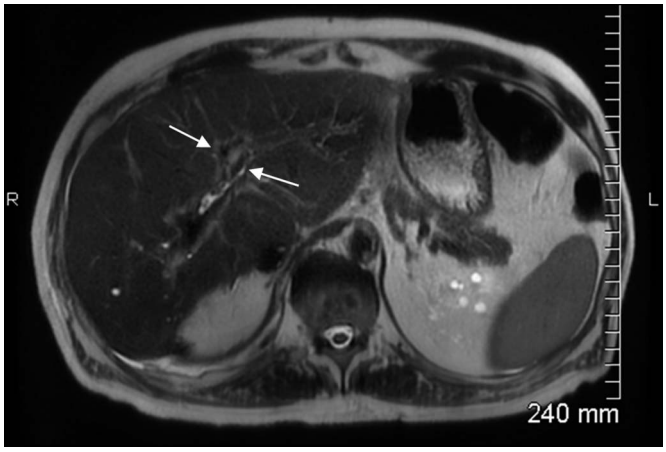

Figure 1. Magnetic resonance imaging before the combined procedure demonstrating left-sided intrahepatic filling defects (arrows directed at filling defects).

cholangioscopy was then performed with a single-operator cholangioscope (SpyGlass 2; Boston Scientific, Boston, MA) passed through a 14 French transhepatic sheath (Figures 3 and 4). Using the cholangioscope, we were able to visualize impacted stones within the left intrahepatic system and treat them using EHL with a catheter setting of 40–50 W for stone fragmentation and suction. The single-operator cholangioscope was then directed at the common bile duct, and 2 large retained stones were treated with EHL. Subsequently, a traditional ERCP was performed which noted filling defects within the common bile duct. Balloon sphincteroplasty was performed and using an 8.5–11.5 mm balloon catheter; CBD exploration and balloon sweeps were performed with extraction of 2 large stones and sludge. An occlusion cholangiogram at the completion of the procedure revealed no further stones and patent anastomosis. Finally, a 14 Fr × 25 cm Dawson Mueller catheter (Cook Medical ; Bloomington) was placed in the common hepatic duct as an external biliary drain, and a retrievable 10 × 60 mm self-

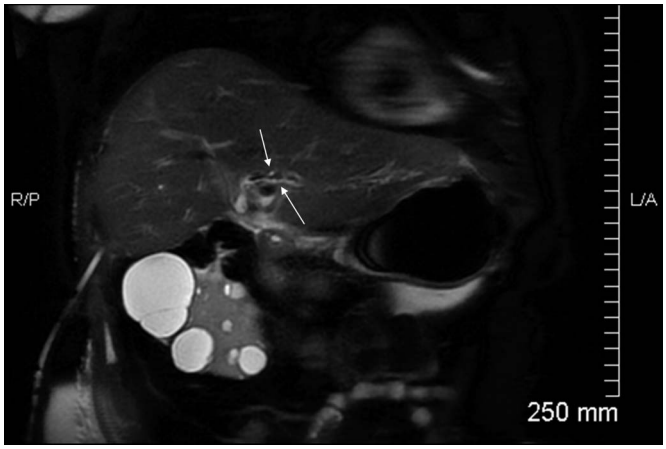

Figure 2. Filling defects noted on a cholangiogram within the left intrahepatic biliary system (arrows directed at filling defects).

Figure 3. Passage of a single-operator cholangioscope through a 14 Fr percutaneous transhepatic sheath.

expanding metal stent-covered metal biliary stent (Viabil; ConMed, Utica, NY) was placed by ERCP (Figure 5).

The total duration of the procedure was approximately 3 hours. The Fogarty balloon was successful in removing about 60% of the stones, and it was easy to pull these from the left-sided bile ducts and push through the CBD. It was not effective for the remaining 40% stones which were adherent to the biliary mucosa and required cholangioscopy for removal. Following this procedure, the dilated tract was managed by serial tube changes downsizing from 14Fr down to 8.5 Fr Dawson Mueller catheter over the next 8 weeks. After a capping trial, external biliary drain was able to be removed 8 weeks after the procedure. The patient was maintained on ursodiol 600 mg AM/300 mg PM after the procedure to prevent further stone formation and did not have any recurrent episodes of cholangitis. The biliary stent was removed 3 months later with ERCP demonstrating no recurrent intrahepatic or extrahepatic bile duct stones. The procedure was

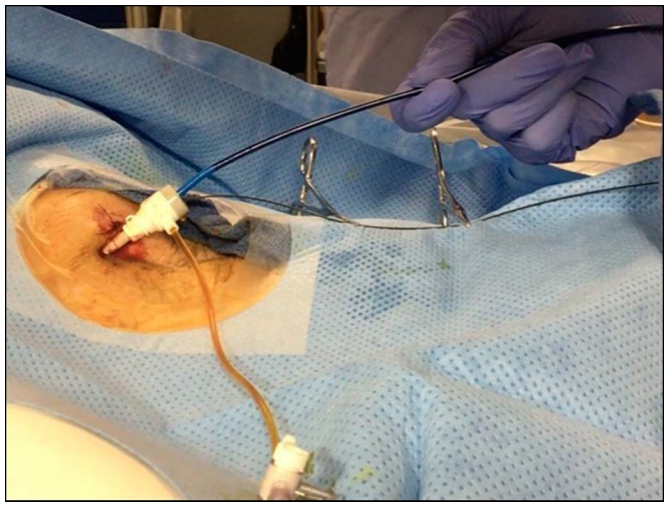

Figure 4. Single-operator cholangioscopy of the left intrahepatic ducts with the direct visualization of stones.

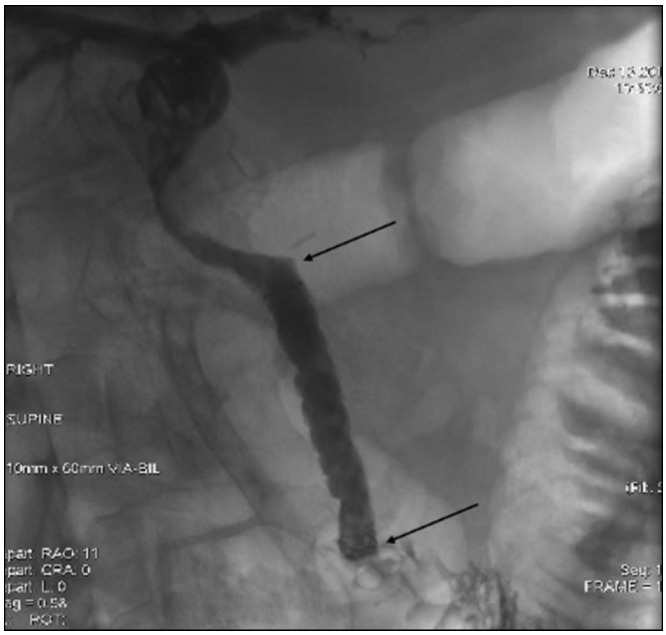

Figure 5. Self-expanding metal stent-covered metal biliary stent placement at endoscopic retrograde cholangiopancreatography (arrows directed at proximal and distal ends).

completed 20 months ago, and most recently, the patient and graft remain well with no further episodes of cholangitis and no drains in place.

DISCUSSION

The blood supply of bile ducts is sensitive to hypoperfusion and places them at high risk for damage in the setting of liver transplantation from both DCD and donation after brain death (DBD) organs. Biliary damage after liver transplantation can manifest as both anastomotic strictures and nonanastomotic strictures. These injuries can lead to the development of sludge, casts, and stones. In a study completed at Pittsburgh over a 13-year period, the prevalence of bile duct-filling defects was 6% after all liver transplants.[1] The formation of sludge, casts, and stones can subsequently result in biliary obstruction, jaundice, cholangitis, and hepatocyte injury.

Intrahepatic cholelithiasis develops in the setting of biliary damage resulting in nonanastomotic strictures. This is seen in the setting of ischemia, which can be secondary to compromised arterial blood supply, prolonged preservation time, or hemodynamic instability surrounding transplantation.[2] Patients, such as the one we discussed, who have received a DCD organ, are at higher risk of developing ischemic cholangiopathy and thus intrahepatic stones and sludge.[3] A meta-analysis published in 2011 demonstrated that DCD recipients had a 10.8 times increased odds of developing ischemic cholangiopathy vs DBD recipients.[3] The same meta-analysis demonstrated that ischemic cholangiopathy was present in 16% of DCD recipients compared with 3% of DBD recipients.[3] Prolonged cold ischemia time in DCD organs seems to be associated with the increased risk of biliary damage.[4] The subsequent development of biliary casts in the setting of DCD organ transplantation has been reported at 35%.[5] A retrospective study demonstrated a risk of recurrent cholangitis of about 25% in patients with non-anastomotic strictures.[6]

With advances in endoscopic retrograde cholangiopancreatography, successful management of common bile duct stones has been made commonplace. Intrahepatic cholelithiasis, however, presents a more difficult challenge. Cannavale et al described an 18-year experience of more difficult to clear intrahepatic cholelithiasis. They demonstrated complete stone clearance with percutaneous transhepatic cholangioscopy and EHL alone in 64.6% of patients. They also demonstrated that through the use of IR techniques in addition to advanced biliary endoscopy resulted in complete stone clearance in 288/299 patients. In their experience, this process could take up to 4 sessions demonstrating the difficult nature of intrahepatic stones.[7] A case report published in 2019 detailed a case in which percutaneous cholangioscopy with laser lithotripsy was used to treat symptomatic cholangiolithiasis in a liver transplant patient with a hepaticojejunal anastomosis stricture.[8] Previous approaches with percutaneous transhepatic cholangioscopy have been traditionally reported with a "daughter" cholangiopancreatoscope or other thin caliber endoscopes which have been used in the technique of percutaneous endoscopic extraction of retained stones.[9] More recently, single-operator cholangioscopy (Spyglass 2) has been used with ERCP to evaluate the biliary system and allow passage of devices such as biopsy forceps, baskets, and EHL probes.[9]

We reported a previous novel case at our institution of percutaneous transhepatic cholangioscopy with EHL in an 89-year-old patient with choledocholithiasis complicating a benign stricture from long-standing placement of an uncovered metal biliary stent. In this case, there was on single large stone impacted within a previous uncovered metal biliary stent causing a distal chronic benign ischemic distal CBD stricture. We were unsuccessful in removal of the stone with a traditional retrograde ERCP route and therefore performed PTC with a subsequent single-operator cholangioscope and EHL, which allowed successful treatment and clearance of the CBD stone.[10] In this second unique case from our institution, we have once again demonstrated the utility of single-operator cholangioscopy system this time in the treatment of complex intrahepatic cholelithiasis using a multidisciplinary collaborative approach between IR and biliary endoscopists. We would consider such an approach in a patient with intrahepatic stones and recurrent infectious complications, including bilomas and cholangitis.

DISCLOSURES

Author contributions: P. Stadmeyer wrote the article. P. Dalvie provided the images. J. Hubers and D. Gopal revised the article for intellectual content. D. Gopal is the article guarantor.

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Informed consent was obtained from the patient for this case report.

Previous presentation: This case was presented at the American College of Gastroenterology Annual Scientific Meeting, October 25-30, 2019; San Antonio, Texas.

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