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Case Report

Cholangiolithiasis postliver transplantation: Successful treatment utilizing percutaneous transhepatic cholangioscopy and laser lithotripsy

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ABSTRACT

Liver transplant is a risk factor for the development of cholangiolithiasis and choledocholithiasis. While usually addressed by endoscopic techniques, percutaneous transhepatic cholangioscopy combined with laser lithotripsy can be considered a suitable alternative option in select patients.

A 29-year-old male with a 27-year history of liver transplant presented with new onset of persistent pain localized to the lower abdomen 9 days after a liver biopsy. Abdominal CT scan and MRCP showed large calculi expanding intra- and extrahepatic bile ducts. All intrahepatic ductal calculi were removed after 2 sessions of laser lithotripsy and basket retrieval, while common hepatic duct calculi were anterogradely swept into the jejunum after balloon cholangioplasty of the hepaticojejunal anastomosis. No major procedure-related complications were seen.

Percutaneous transhepatic cholangioscopy and choledochoscopy with laser lithotripsy is a minimally invasive and efficient technique for removal of intra- and extrahepatic bile duct stones postliver transplantation.

Introduction

Cholangiolithiasis affects between 1.8% and 18% of transplanted livers cases [1,2], and is usually a sequela of hepaticojejunal anastomosis (HJA) strictures. Left untreated, biliary stones can lead to biliary obstruction, cholestasis, infection, as well as ductal and hepatocyte damage, leading to cholestatic cirrhosis with eventual liver failure or graft loss [3,4].

Reconstructive surgery for strictures of HJA and cholangiolithiasis results in a decreased quality of life and an increased risk of developing recurrent strictures [5]. Percutaneous

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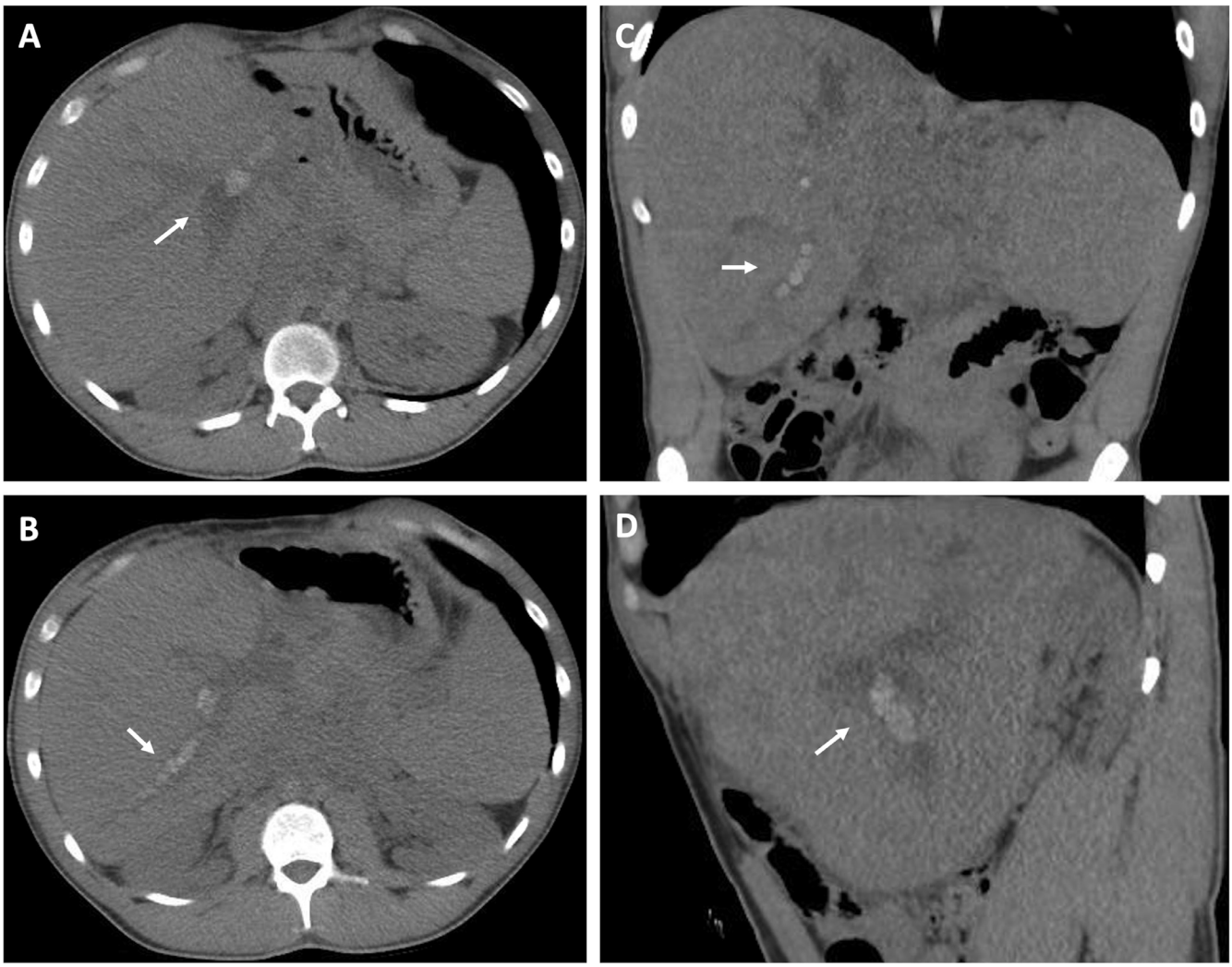

Fig. 1 – CT abdomen and pelvic on the day of presentation. Axial (A and B), coronal (C), and sagittal (D) views. Extensive hyperdensities (white arrows) within tubular structures on this noncontrast CT in this post liver transplant and recent postliver biopsy may represent; blood in the bile duct, biliary calculi or possibly venous thrombosis.

transhepatic endoscopy is an alternative to surgical or retrograde endoscopic treatment of bile duct stones [6,7]. This technique provides a safe and effective approach for the treatment of not only extrahepatic, but also intrahepatic bile stones [7]. Herein, we present the case of a liver transplant patient with a large burden of biliary stones within the intra- and extrahepatic bile ducts, who was successfully treated using percutaneous transhepatic cholangioscopy/choledochoscopy in combination with laser/mechanical lithotripsy and basket stone retrieval.

Case presentation

A 29-year-old male with a history of orthotopic liver transplant, in the setting of biliary atresia at 19 months of age, presented to the emergency department 9 days after liver biopsy for evaluation of abdominal pain. The liver biopsy was performed because of elevated liver function tests after noncompliance with his immunosuppression. The patient reported new onset of constant right upper quadrant abdominal pain starting the morning of presentation with severity of 5/10, without modifying factors. A noncontrast CT abdomen and pelvis (Fig. 1A–D) in the emergency department demonstrated hepatomegaly with multiple varying size hyperdensities within the dilated intrahepatic bile ducts. The following day, magnetic resonance cholangiopancreatography (Fig. 2A–D) indicated multiple stones within the extensively dilated intrahepatic bile ducts (Fig. 2A and B) most prominent in the posterior right bile ducts but present throughout. A centrally situated stone measuring approximately 1 cm was noted in the right main duct (Fig. 2C).

Endoscopic retrograde cholangiopancreatography (ERCP) on the same day failed to access the biliary roux-limb and unfortunately was complicated by pancreatitis. Given the degree of intrahepatic biliary dilatation and ongoing cholangitis picture, interventional radiology was consulted for a percutaneous biliary drain placement.

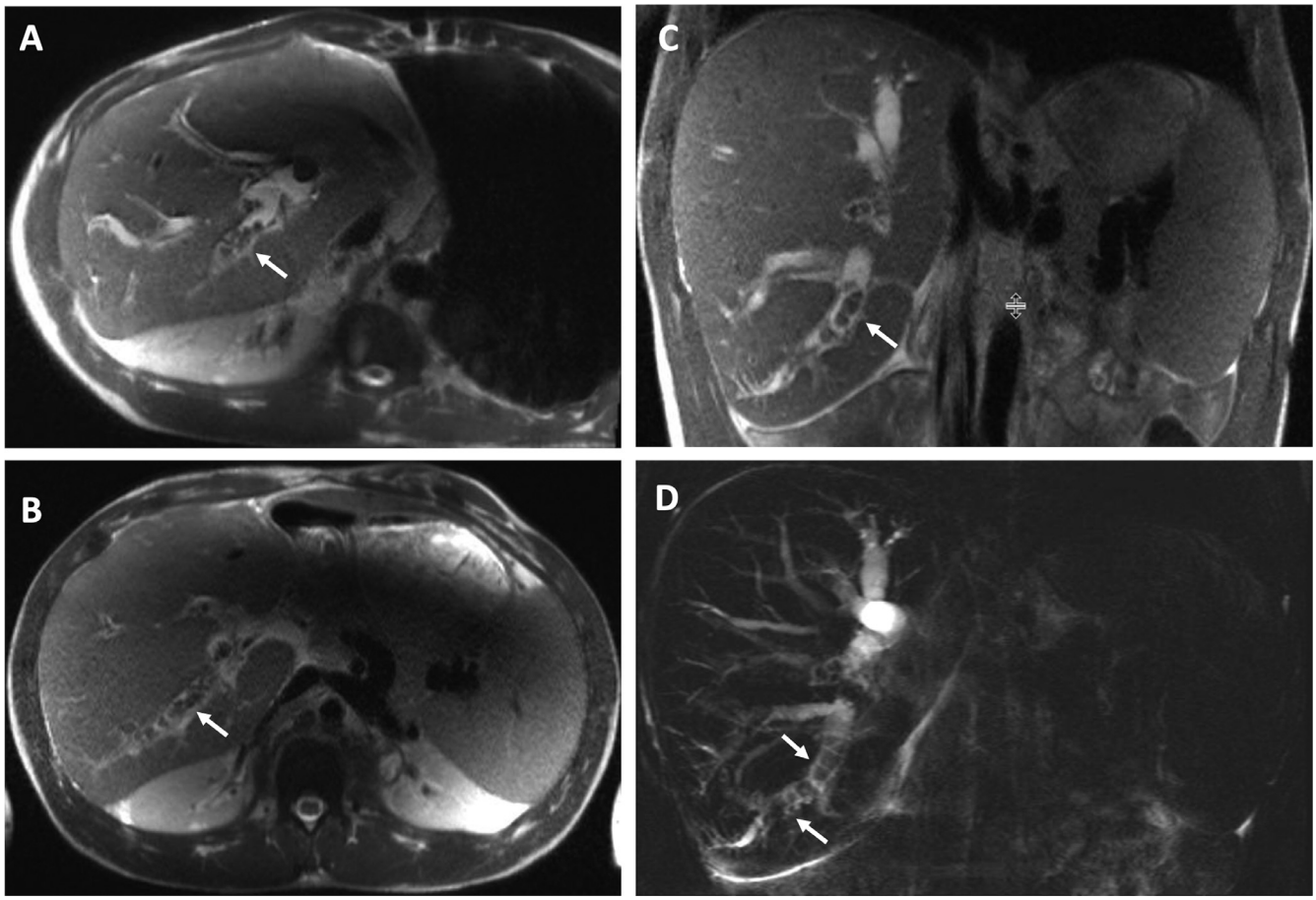

Fig. 2 – MRI of abdomen 1 day after presentation. Axial (A and B), coronal (C), and MRCP (D) views. Liver transplant is noted in the right upper quadrant. There is marked intrahepatic biliary dilatation, with multiple intraluminal gallstones (white arrows) including a 1-cm stone at the right main duct and near complete occlusion of the right posterior biliary duct.

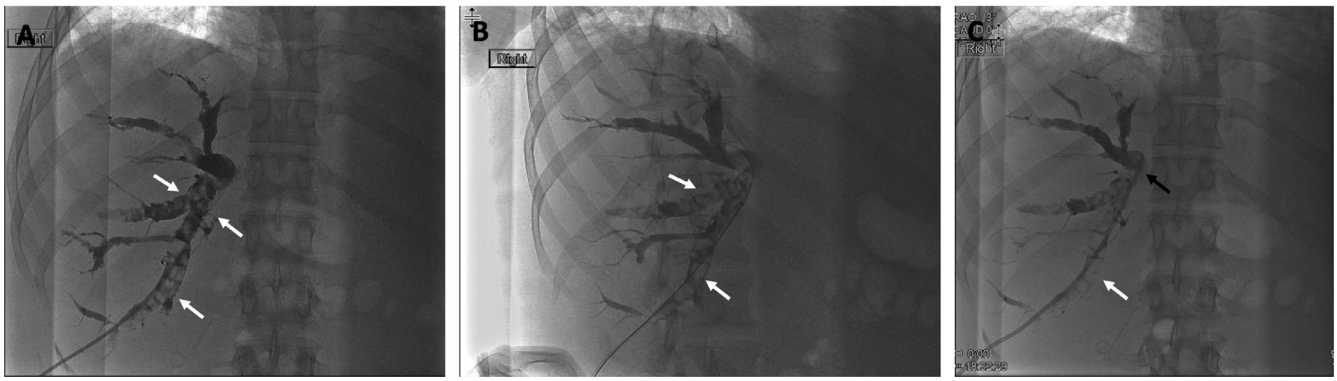

Fig. 3 – Preliminary cholangiogram and biliary drainage catheter placement 5 days after presentation. (A) and (B). Frontal and right anterior oblique views showed diffuse dilatation of the opacified right biliary ducts with numerous filling defects (white arrows) concordant with biliary stones. (C) Placement of a right-sided 8 French external biliary drainage catheter (black arrow) via subcostal midaxillary approach with the loop formed near the confluence of the right hepatic ducts.

Initially, a right percutaneous transhepatic cholangiogram was performed that opacified the right biliary ducts and revealed diffuse dilatation of intra- and extrahepatic bile ducts with numerous filling defects throughout—most consistent with biliary calculi (Fig. 3A and B). Given the purulent bilious output, an 8 French external pigtail catheter with extra proximal side holes was placed via a subcostal midaxillary approach to minimize biliary manipulation (Fig. 3C). The pigtail part of the catheter was positioned near the confluence of the right hepatic ducts, and side

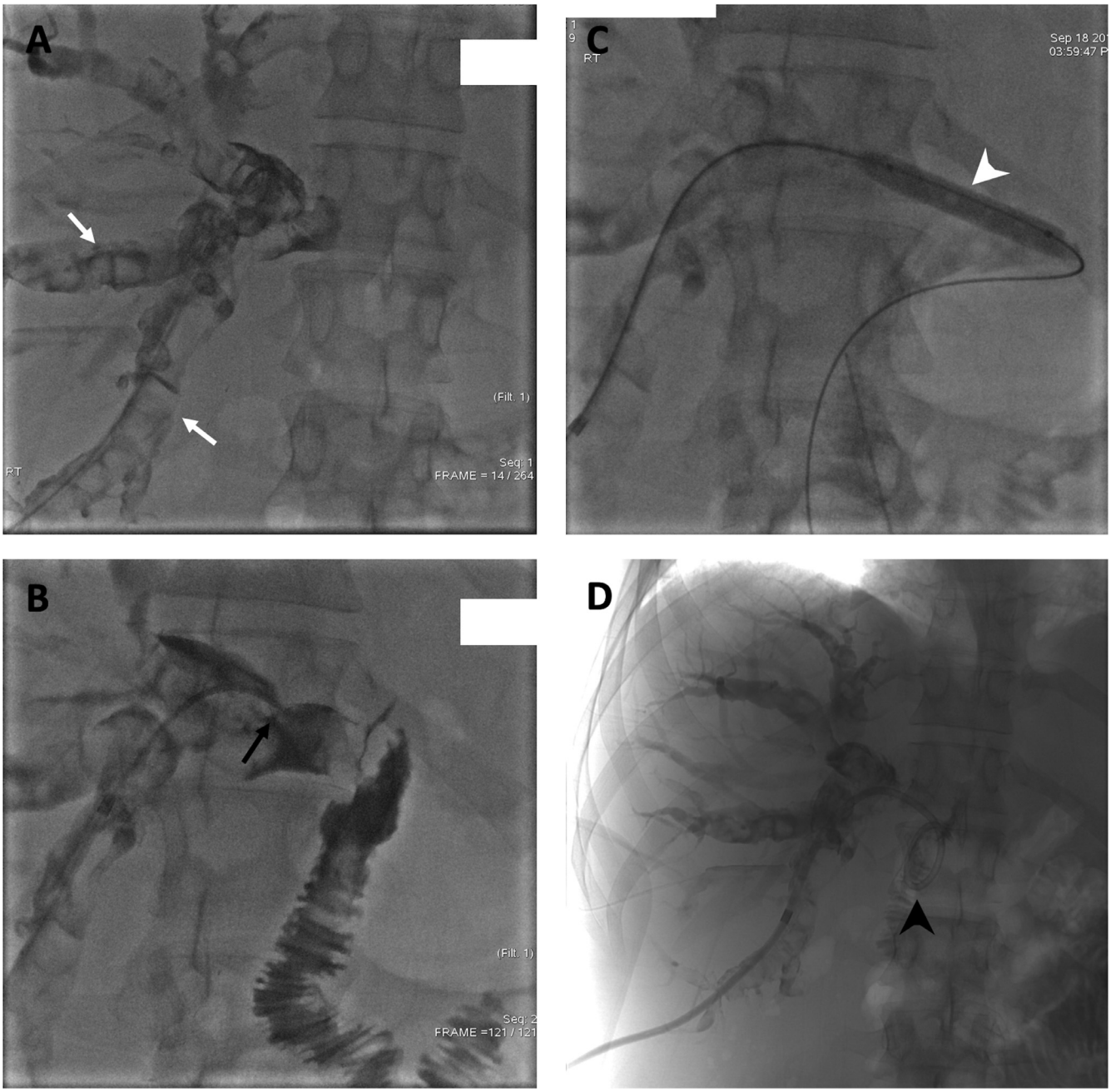

Fig. 4 – Follow-up over-the-wire cholangiogram 12 days after initial cholangiogram. (A) Cholangiogram revealed numerous filling defects representing intraductal stones (white arrows) in the right intrahepatic biliary system. (B) There was a large common bile duct stone at the stenotic hepaticojejunostomy anastomosis (black arrow). (C) An 8-mm Mustang Balloon (white arrow head) was used in to sweep stones from the central ducts antegrade into the jejunum. Balloon dilation of the hepaticojejunal anastomosis was performed with 10 mm and 12-mm balloons. (D) A new 12 French biliary drainage catheter was advanced over the wire into the biliary tree. The distal pigtail was formed in the duodenum (black arrow head).

holes were aligned along the anterior-inferior right hepatic duct.

Follow-up over-the-wire cholangiogram, 1 week after initial access, revealed numerous filling defects representing intraductal calculi throughout the right intrahepatic biliary system (Fig. 4A), and a large common bile duct stone at the stenotic HJA (Fig. 4B). Balloon dilation of the HJA was accomplished using 10 and then 12-mm balloons (Fig. 4C). Then, an 8-mm Mustang Balloon was used in anterograde fashion to sweep stones from the central ducts into the jejunum. Subsequently, a new 12 French internal-external biliary drainage catheter was placed (Fig. 4D).

At 2 weeks follow-up, the internal-external biliary drainage catheter was exchanged for a 14 French vascular sheath, and

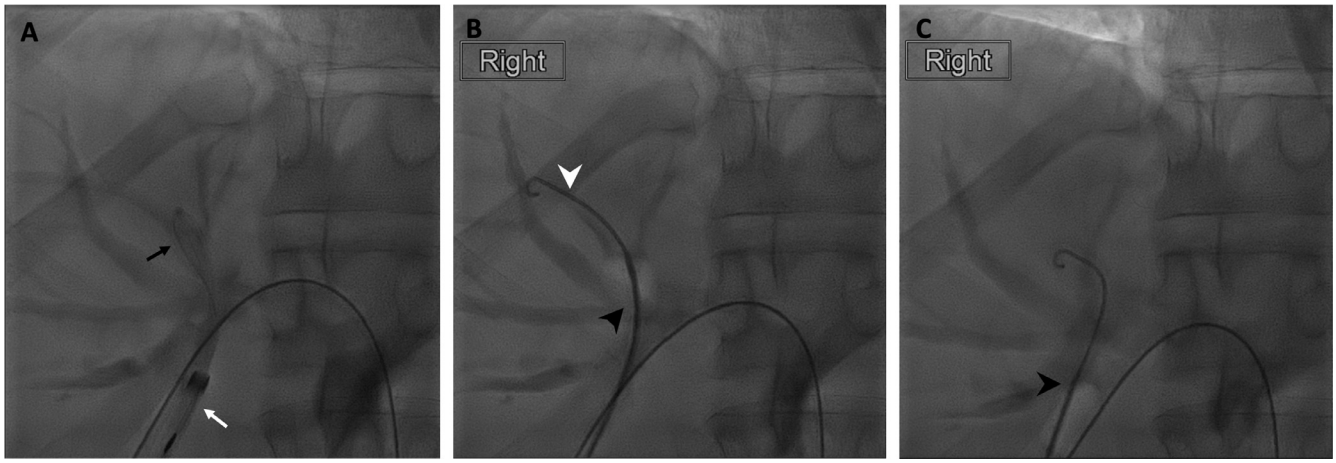

Fig. 5 – Cholangiogram approximately 5 weeks after presentation. (A) A 14 French sheath (white arrow) was positioned at the entry site to the accessed right biliary duct. Subsequently, a Wittich nitinol basket (black arrow) was applied to extract a large amount of stones from the accessed right biliary duct. (B) A 5.5 French Fogarty balloon (white arrow head) was used for sweeping of a large stone out of a more superior duct (black arrow head). (C) However, the stone (black arrow head) was too large to be retrieved with a basket or swept beyond the hepaticojejunostomy.

stone retrieval with a Wittich nitinol basket was performed. This extracted a large amount of stones from the accessed right biliary system (Fig.5A). A 5.5 French Fogarty balloon was then used to attempt sweeping a stone out of a more superior duct (Fig. 5B), but the stone was too large to be displaced with a basket or swept beyond the HJA (Fig. 5C).

During the subsequent session, a 9.5 Fr LithoVue scope (Boston Scientific, Marlborough, MA) was introduced into the biliary system through the 14 French sheath. Visualization of the filling defects confirmed them as calculi. A 5.5 French Fogarty balloon was then used to sweep multiple stones from the peripheral duct to the main ducts where they were retrieved by a basket (Fig. 6). Subsequently, several biliary stones were fragmented using Lumenis Pulse 30H Holmium laser (Lumenis Lts., Yokneam, Israel) and Felxiva laser fiber of 365 nm (Boston Scientific, Salem, NH) with a laser frequency of 12 Hz and 1.2 J energy under direct visualization. Completion cholangioscopy and cholangiogram demonstrated minimal residual large biliary stones in the intrahepatic biliary system. Additionally, there was a persistent focal narrowing in the central segment of the right biliary duct (Fig. 7A). Therefore, a 12 French drainage catheter was advanced over the wire and positioned with the proximal side holes in the central to midright biliary system. An additional session of laser lithotripsy and basket retrieval under cholangioscopic guidance was performed 3 weeks later. At its completion, a new 18 French Heyer-Schulte biliary tube was manipulated into the biliary system such that the proximal side holes were within the biliary tree, and no major area of the biliary ductal system was left undrained.

Two weeks later, a cholangiogram through the biliary drain showed no filling defects (Fig. 7B). Overall, 5 sessions were performed during a period of 3 months to clear all visible biliary stones—the prolonged time frame was at request of the patient. The catheter was removed a week later following a successful external only biliary drain capping trial. Follow-up magnetic resonance cholangiopancreatography of abdomen 5 months later showed no biliary system dilatation or residual stones (Fig. 8).

Discussion

We present a case of postliver transplantation cholangiolithiasis that was successfully treated with percutaneous transhepatic cholangioscopy/choledochoscopy in combination with laser lithotripsy, retrograde basket stone removal, and anterograde balloon-assisted stone clearance. All biliary calculi were successfully cleared and the HJA stricture dilated after multiple sessions of treatment and the biliary drainage catheter was eventually removed.

Cholangiolithiasis includes debris, sludge, casts, and stones within the intrahepatic bile ducts and can affect between 1.8% and 18% of transplant liver recipients [1,8]. If untreated, this condition could result in biliary obstruction, cholestasis, infection, as well as ductal and hepatocyte damage, leading to cholestatic cirrhosis with eventual liver failure or graft loss [3].

Although the etiology of cholangiolithiasis is still unclear, there are reports relating this finding to environmental factors, nutritional status, bile duct infection, cholestasis, parasites, variation of bile ducts, and bile metabolic defects [9]. Cholestasis, however, is one of the most important and common causes of biliary stone formation. Postliver transplantation, cholestasis can affect extrahepatic bile ducts, due to mechanical obstruction of the main bile ducts, or intrahepatic bile ducts, due to impairment of bile duct secretion in the setting of malfunction of the hepatocytes or microscopic intrahepatic bile ducts [10]. Additionally proposed etiologies of transplant-related intrahepatic cholestasis include inflammatory changes or destruction of bile ducts, bacterial or viral infection, hepatotoxic medications such as

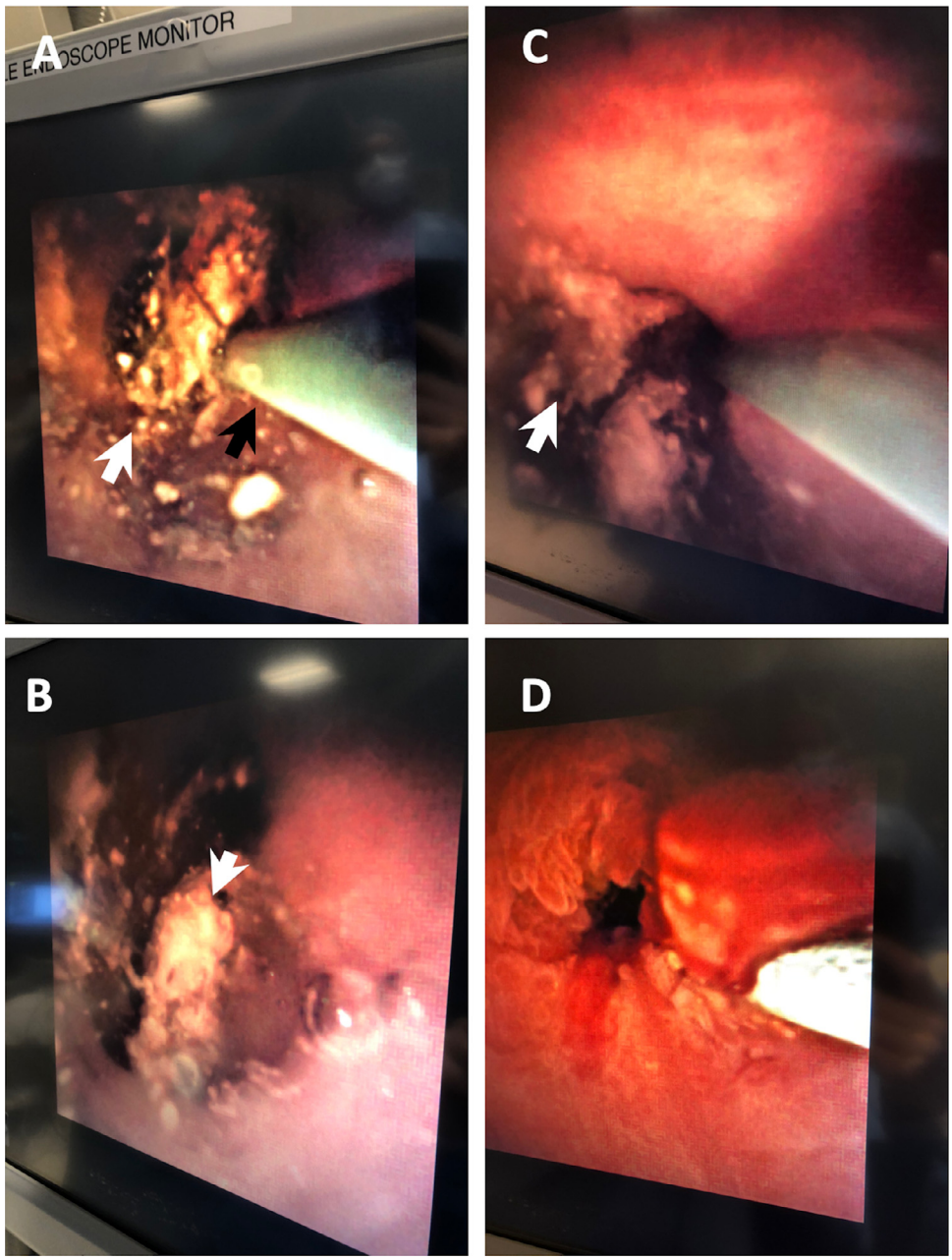

Fig. 6 – Cholangioscopy 8 weeks after first presentation. (A-D) Eight weeks after the last cholangiogram, laser (black arrow) and mechanical lithotripsies were used under cholangioscopic guidance with Fogarty balloon to remove large hilar stone (white arrow) and other multiple stones.

immunosuppressive drugs including azathioprine and sulfonamides, ischemic/reperfusion injury, or a combination of these factors [11].

The diagnosis of biliary stones is based on imaging and a variety of treatment options have been used. Surgical treatment mainly includes bile duct incision and lithotomy, hepatic resection, reconstruction of bile duct stricture, and in extreme cases liver retransplantation. While surgical approaches used to be the cornerstone of treatment, there has been a transition from surgical treatment approaches to minimally invasive procedures for the diagnosis and treatment of cholangiolithiasis and biliary system strictures. These can be divided into retrograde and antegrade approaches. Retrograde endoscopy (ERCP) is typically performed by a gastroenterologist and it includes transoral passage of an endoscope with eventual cannulation of the bile duct. Direct cholangioscopy is typically performed by an interventional radiologist and it includes percutaneous transhepatic biliary drainage, cholangiography, and cholangioscopy. Both approaches can involve balloon dilatation of the HJA, lithotripsy with basket or balloon assisted stone extraction, and stenting [7,12].

The efficacy of percutaneous transhepatic interventions for strictures of HJA and associated cholangiolithiasis ranges from 67% to 73% [13,14]. A recent study reported 22.4%

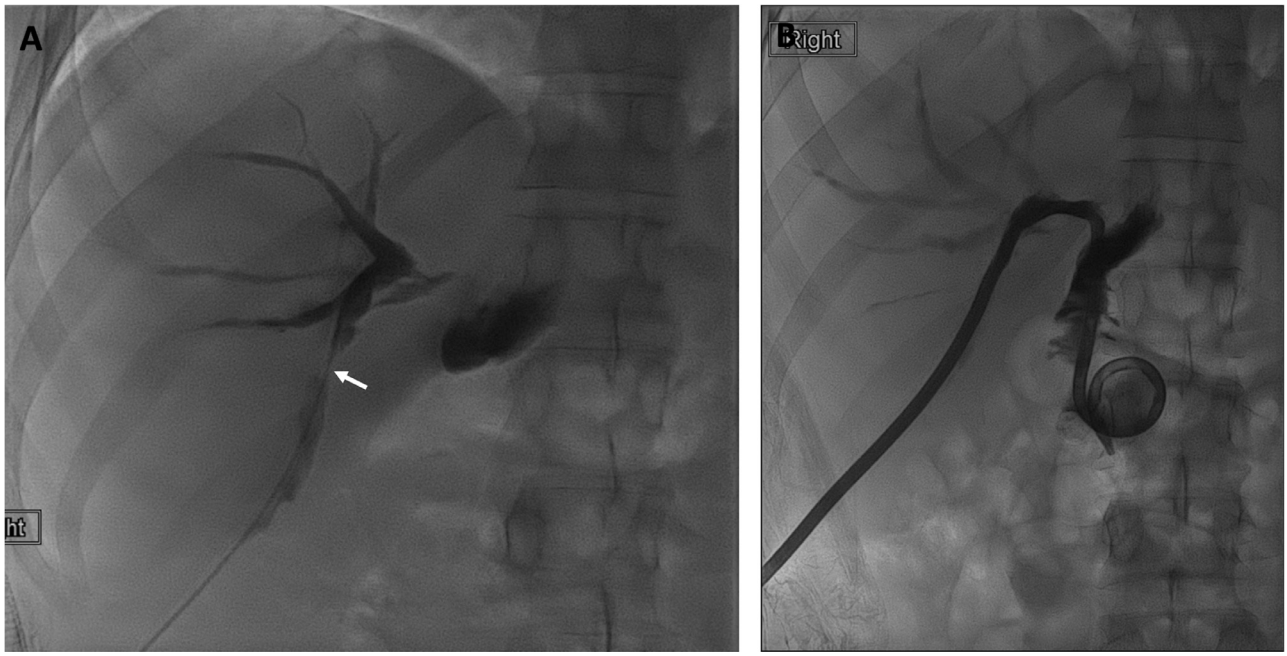

Fig. 7 – Cholangiogram approximately 3.5 months after presentation. (A) Cholangiogram showed persistent focal narrowing (white arrow) in the central segment of the right biliary duct. (B) Cholangiogram through the 12 French biliary drain showed no definite filling defects.

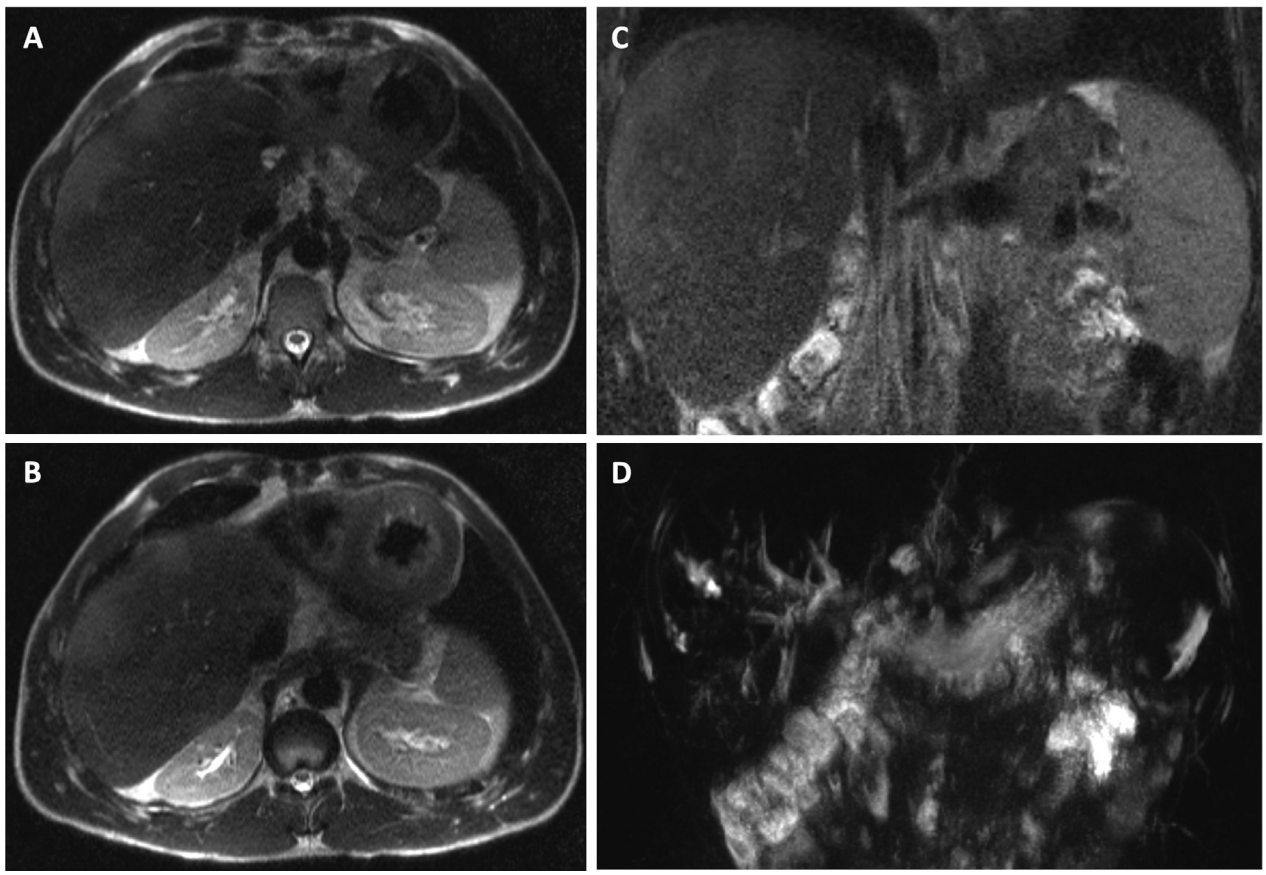

Fig. 8 – MRI of abdomen 6 months after removal of biliary drains. Axial (A and B), coronal (C), and MRCP (D) views. Liver transplant is noted in the right upper quadrant. There is no intrahepatic biliary dilatation or intraluminal gallstones.

intrahepatic cholangiolithiasis within 1 to 3 years after reconstructive surgery of HJA strictures, and 100% successful treatment rate with minimally invasive procedures performed on 20.7% of these patients. A judicious treatment algorithm allows for not only identification of the HJA stricture and intra- or extrahepatic biliary stones, but also restores biliary drainage by minimally invasive means.

Conclusion

The combination of antegrade balloon cholangioplasty of HJA stricture, cholangioscopy or choledochoscopy, laser and mechanical lithotripsy with retrograde basket and anterograde balloon assisted stone extraction presented here are an efficacious and safe approach to treat strictures of HJA and cholangiolithiasis post hepatic transplantation.

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