

CASE REPORT

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Successful application of percutaneous endoscopic cholangioscopy + electrohydraulic lithotripsy for hepatolithiasis post-liver transplantation: a case report

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Abstract

Background Liver transplantation in pediatric patients is a crucial intervention for treating end-stage hepatic diseases. Despite significant advances in surgical techniques and postoperative care, complications remain a substantial challenge in this population. Biliary stones, an infrequent complication, present challenges in this context. Given the impossibility of endoscopic treatments, different strategies have been explored to address post-liver transplantation gallstones in children by implementing percutaneous treatment with intraductal lithotripsy.

Case presentation A seven-year-old Latin patient who had a diagnosed at biliary atresia, at the age of two underwent a liver transplant from a living donor. However, four months after the transplant, the patient experienced recurring episodes of cholangitis. Cholangioresonance revealed intrahepatic lithiasis and anastomotic stenosis. Attempted gallstone removal through percutaneous cholangiography proved unsuccessful, as multiple peripheral stones in all ducts remained immobile. Subsequently, a percutaneous endoscopic cholangioscopy using the SpyGlass Discover system, for visual examination of the bile ducts + electrohydraulic lithotripsy was performed, effectively removing the stones without any complications.

Conclusions Percutaneous cholangioscopy with intraductal lithotripsy enables accurate identification and extraction of intrahepatic stones without the need for surgical intervention. This method proves to be a valuable alternative in addressing post-transplant biliary stone. In our case, it was performed on a pediatric patient with liver transplantation, which makes it interesting and relevant as there is currently insufficient literature on this approach in such cases in this population.

Keywords Percutaneous cholangioscopy, Spyglass, Liver, Lithotripsy

Background

Post-transplant biliary complications remain a significant concern, with a global incidence ranging from 15 to 40% in pediatric transplant recipients [1]. Biliary stones, present in 2–6% of liver transplant recipients, often result from anastomotic stenoses and are associated with severe complications [2]. Post-transplant cholestasis, attributed to genetic factors, elevated lipid levels, increased inflammatory response, and ischemia/reperfusion, contributes to biliary sludge formation. While surgical approaches

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were once prioritized, endoscopic procedures have proven to be preferable in the long term [3]. However, in cases where endoscopic cannulation fails due to anatomical variations, percutaneous endoscopic cholangioscopy emerges as a safe and effective alternative. We present a successful case of managing intra and extrahepatic bile duct stones in a pediatric patient with a history of liver transplantation using percutaneous cholangioscopy combined with laser/mechanical lithotripsy.

Case presentation

A seven-year-old Latin patient with a history of Kasai surgery (portoenterostomy) for extrahepatic biliary atresia at the age of two underwent a liver transplant from a living donor at eight months due to a failed initial Kasai procedure, resulting in a Roux-en-Y hepaticojejunostomy. However, four months post-transplant, the patient experienced recurrent episodes of cholangitis, prompting a Doppler ultrasound revealing significant stenosis of the portal anastomosis. Addressing this complication required three percutaneous transhepatic biliary drainages (PTBD) and replacements of biliary diversion catheters in an attempt to correct the bilioenteric stenosis ranging from 30 to 60%.

During the latest emergency admission, the patient was diagnosed with a new episode of ascending cholangitis, accompanied by intrahepatic lithiasis and anastomotic stenosis revealed by cholangioresonance (Fig. 1). Subsequent fluoroscopic and ultrasound review indicated intrahepatic bile duct dilation with stenosis at the anastomosis and the presence of stones. A 10 cm × 4 cm

balloon dilation was performed with successful cholangiographic results, partially clearing some stones into the intestine. However, multiple peripheral stones in all ducts remained immobile, necessitating the placement of an 8 Fr internal–external biliary catheter.

However, the patient returns to the emergency department one month after the procedure due to itching sensation and dysfunction of the drain. Subsequently, a percutaneous endoscopic cholangioscopy + lithotripsy is performed.

During the procedure, a previously inserted 10 Fr percutaneous catheter placed by Interventional Radiology was used to introduce a cholangioscope (SpyGlass Discover) into the left intrahepatic bile duct. Three yellow pigment stones with a hard consistency and a diameter of approximately 5–6 mm were observed. Additionally, an inflammatory stenosis was identified at the hepatojejunal anastomosis (Fig. 2). To address the stones, electrohydraulic lithotripsy was performed at a medium power of 10 and 200 shots, effectively fragmenting the stones (Fig. 3). Subsequently, the stones were advanced into the small intestine through the anastomosis without complications. A hydrophilic guide was left in the jejunal loop for continued intervention, and radiology intervention placed a stent, leaving a functioning drain (Fig. 4). At the end of the procedure, an 8Fr internal–external drainage was left in place.

Follow-up: Three months post-procedure, cholangiography is performed through the catheter, showing good contrast passage into the intestine with no evidence of stenosis or stones. The catheter and stent were removed (Fig. 5).

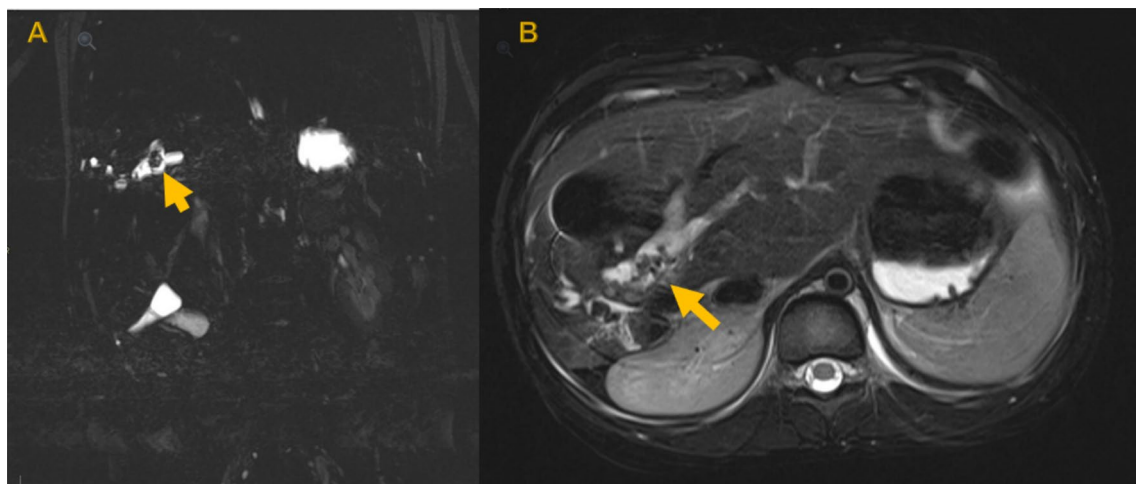


Fig. 1 MR Cholangiopancreatography. **A** Coronal view: Biliary stones are observed (yellow arrow). **B** Axial view: Biliary stones are visualized (yellow arrow)

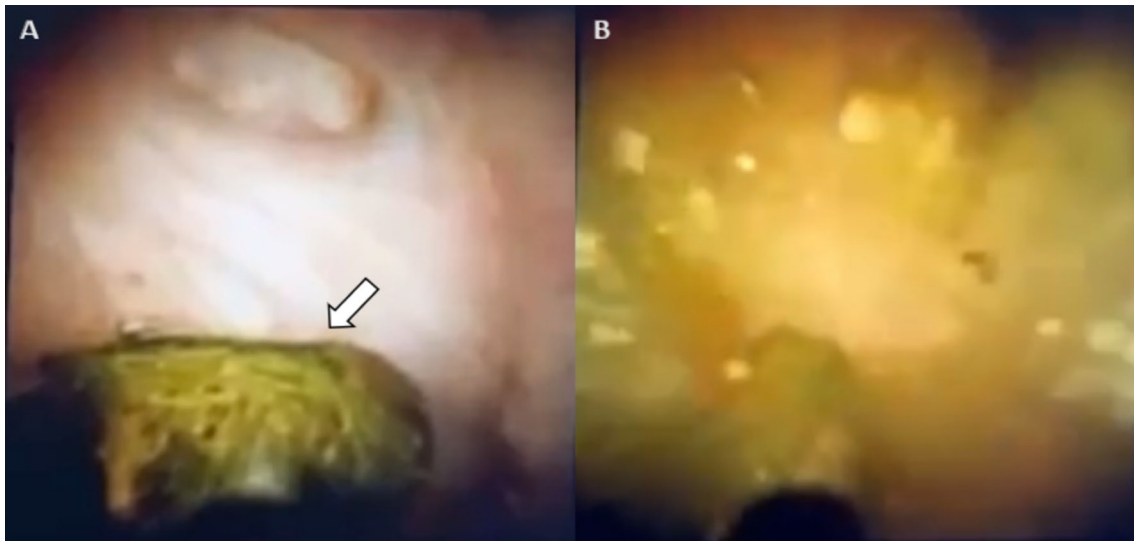


Fig. 2 Percutaneous endoscopic cholangioscopy + electrohydraulic lithotripsy. **A** Visualization of biliary stone (White arrow). **B** Biliary stone fragments after procedure

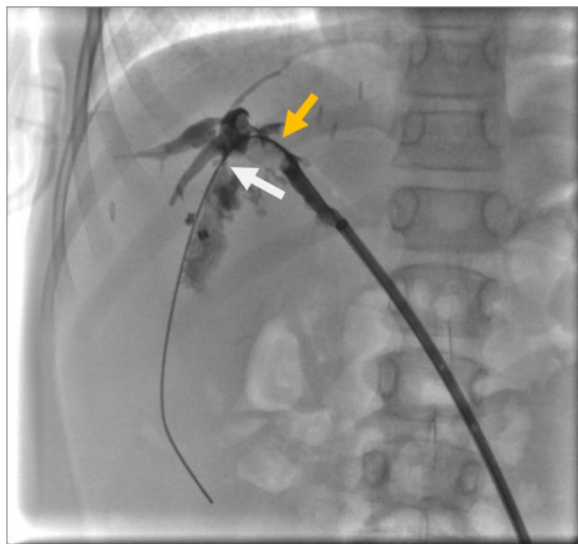


Fig. 3 Biliary cholangiography. The yellow arrow reveals the presence of a bile duct stone. The white arrow reveals the presence of bilioenteric anastomotic stenosis



Fig. 4 Biliary cholangiography. White arrow: Placement of a removable percutaneous biliary stent is observed to resolve the stenosis. Yellow arrow: Following stent placement, the disappearance of the stone is also evident

Discussion

We present a case of a 7-year-old pediatric patient with post-liver transplant hepatolithiasis successfully treated through percutaneous transhepatic cholangioscopy in combination with laser lithotripsy. The procedure included prior balloon dilation of the hepatojejunal anastomosis and subsequent removal of the biliary drainage catheter without complications.

The formation of biliary stones after liver transplantation represents a significant clinical challenge, as it not

only entails risks of morbidity and mortality but can also lead to recurrent episodes of cholangitis and, ultimately, trigger secondary biliary cirrhosis [4]. Diverse factors, whether inflammatory, physical, or metabolic, heighten the susceptibility to biliary stones formation, with the reduction of bile flow emerging as the primary determining factor [5]. In patients subjected to liver transplantation, cholestasis emerges as one of the most prevalent and impactful causes of postoperative biliary stones development [6]. This condition affects both extrahepatic and intrahepatic bile ducts, resulting from either mechanical obstructions or disruptions in secretion. Other potential

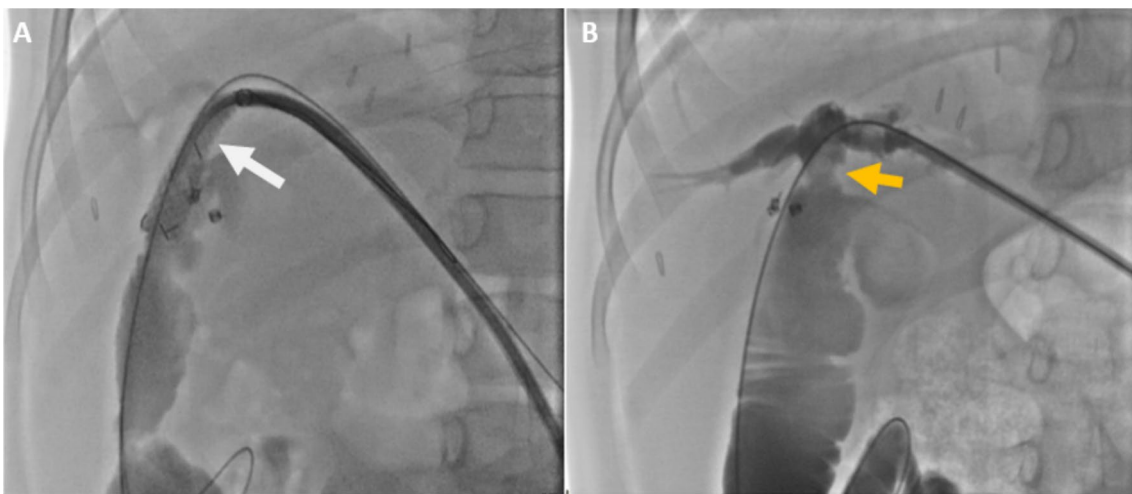


Fig. 5. 3-month follow-up by cholangiography. **A** White arrow: Removable percutaneous biliary stent. **B** Yellow arrow: Post-removal of the biliary stent, where a normal bilioenteric anastomosis is visualized

causes associated with intrahepatic cholestasis after transplantation include inflammatory changes or the loss of integrity of bile ducts, bacterial or viral infections, the administration of hepatotoxic drugs such as immunosuppressants, as well as injuries resulting from ischemia/reperfusion processes or a combined interaction of these factors [7].

Diverse therapeutic strategies have been employed to address the formation of biliary stones in patients undergoing liver transplantation. Despite the previous prioritization of surgical approaches as the cornerstone of treatment, there has been a transition towards minimally invasive procedures for the diagnosis and treatment of biliary system lithiasis and stenosis. Although endoscopic procedures, such as endoscopic retrograde cholangiopancreatography (ERCP), generally achieve a definitive treatment with a success rate exceeding 90%, it is essential to recognize that not all cases respond efficiently to these techniques [8].

Currently, post-transplant biliary complications have been primarily addressed through endoscopic and percutaneous interventional procedures, as they offer safety, minimal invasiveness, and favorable outcomes compared to surgical management [8].

In cases of complex lithiasis, marked by the presence of numerous, sizable, intrahepatic stones, or those situated in regions with anastomotic stricture, advanced intraductal techniques such as electrohydraulic lithotripsy using SpyGlass cholangioscope, a single-operator endoscope, have demonstrated exceptional results [8]. This therapeutic modality provides the ability to fragment stones without significantly compromising biliary integrity, addressing situations that go beyond the scope

of conventional endoscopic techniques. These strategies are particularly relevant in challenging cases that do not respond adequately to standard ERCP treatment, and where lithiasis persists despite previous attempts [8]. A review of multiple studies on cholangioscopy revealed the occurrence of potential side effects such as pancreatitis, cholangitis, and perforation. However, there is limited information on device-related failures or patient-associated events related to the SpyGlass cholangioscope [9].

In our specific case, the patient had previously undergone a cholangiography, where, unfortunately, the complete removal of the stones was not achieved, revealing the persistence of multiple intrahepatic stones. This scenario underscores the necessity of tailoring therapeutic strategies to the individual complexity of each case, acknowledging that endoscopic techniques may not always be sufficient, and alternative options can be crucial to achieve successful outcomes.

In conclusion, the combination of a percutaneous approach with cholangioscopy and lithotripsy emerges as a viable and safe option for managing biliary complications in pediatric patients with a history of liver transplant. This is particularly pertinent as literature on such cases is limited. Moreover, it paves the way for further research into this procedure and other potential interventions within this cohort of pediatric liver transplant recipients. Although additional studies are needed to validate its effectiveness specifically in this population, the presented case underscores the importance of a multidisciplinary approach and collaboration among specialists. This comprehensive approach proves to be a key factor in improving outcomes in complex clinical situations, emphasizing the significance of ongoing research and

knowledge exchange in the care of patients with a history of liver transplant and biliary lithiasis.

Conclusion

The percutaneous approach with cholangioscopy plus lithotripsy may be a viable and safe option for managing biliary complications in pediatric patients with a history of Kasai surgery and liver transplantation. The limited literature available in this area emphasizes the need for further research on this procedure and other possible interventions. However, further studies and case reports are needed to validate its effectiveness in this specific population. This case illustrates the importance of a multidisciplinary approach and collaboration among specialists to enhance outcomes in complex clinical situations.

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Author contributions

AC: project administration, review and editing. DN: conceptualization—writing—original draft. JT: conceptualization and review. AH: conceptualization and review.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

The reported case was reviewed and approved, and individual patient consent was obtained following institutional guidelines. Following our institutional policies, all protected health information was removed.

Consent for publication

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare that they have no competing interests.

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