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Case Report

Percutaneous transhepatic biliary intervention for the management of recurrent cholangitis secondary to intrahepatic biliary stones after hepaticojejunostomy[☆]

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ABSTRACT

Recurrent cholangitis, intrahepatic stones and biliary and anastomotic strictures are common complications after Roux-en-Y hepaticojejunostomy. The surgically altered anatomy makes management of these complications with endoscopic retrograde cholangiopancreatography technically difficult. We present a case of recurrent cholangitis in a 25-year-old woman with a prior hepaticojejunostomy. Intrahepatic stones, biliary strictures and hepaticojejunostomy strictures were treated using a combined percutaneous transhepatic cholangiography and cholangioscopy approach over several interventions. Minimally invasive, multidisciplinary approach to the management of intrahepatic biliary calculi and stricture after hepaticojejunostomy can be considered in cases where traditional endoscopic retrograde cholangiopancreatography intervention is too technically difficult.

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Introduction

Bile duct resection and reconstruction is performed for congenital abnormalities in approximately 2.5% of cases [1]. Choledochal cysts are congenital dilatations of the biliary tree. There are 5 subtypes based on the type and location of dilata-

tions. Type II is a biliary tract diverticulum, commonly arising from the common bile duct (CBD) [2]. Aims of surgical management are to mitigate malignancy risk and prevent complications. This is most commonly accomplished with total cyst excision and Roux-en-Y hepaticojejunostomy formation [3,4].

Complications following hepaticojejunostomy are common and include intra-abdominal collections, bile leaks,

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wound infection, anastomotic stricture (11%), spontaneous bacterial cholangitis, recurrent cholangitis (7%–14%), hernias, duodenal ulceration, and hepatolithiasis (2.5%) [5,6]. Rates of biliary stricture are high (57.8%) among patients who develop cholangitis after hepaticojejunostomy [5].

Acute cholangitis and biliary sepsis are life-threatening complications. Following hepaticojejunostomy, biliary obstruction and cholangitis may result from intrahepatic duct stricture, anastomotic stricture, hepatolithiasis or a combination. Endoscopic retrograde cholangiopancreatography (ERCP) is the treatment of choice to establish biliary drainage [7]. Therapeutic manoeuvres include stone extraction, balloon dilatation, and stent insertion. In patients with Roux-en-Y anatomy, traditional ERCP is challenging due to the length and angulation of bowel that must be traversed [8]. When not technically feasible, alternatives via transoral, surgical or percutaneous approach are considered on a case-by-case basis.

Percutaneous transhepatic cholangiography (PTC) is performed via transhepatic needle insertion into a bile duct, followed by contrast material injection to opacify and visualize the ducts. It is an alternative method to facilitate therapeutic intervention and management of biliary obstruction in patients with surgically altered anatomy [9]. Following percutaneous biliary access, percutaneous transhepatic cholangioscopy using a miniature intraductal endoscope permits direct visualization of the biliary tree and an additional approach to management of intrahepatic calculi [10].

The following case outlines a multidisciplinary, minimally invasive approach to managing recurrent cholangitis following hepaticojejunostomy. The patient provided written consent for the publication of this report and ethics approval was received from the institutional ethics committee.

Case report

We present the case of a 25-year-old patient, who was managed for biliary calculi and strictures after a previous hepaticojejunostomy with percutaneous transhepatic biliary intervention. Her past medical history was otherwise significant for obesity and asthma. There was no significant family history.

The patient initially presented with choledocholithiasis. Magnetic resonance cholangiopancreatography (MRCP) identified a type II choledochal cyst which was managed with laparotomy, choledochal cyst excision, cholecystectomy and Roux-En-Y hepaticojejunostomy. Following this, they were admitted 5 times for recurrent cholangitis over several years, each episode managed with intravenous antibiotics. Subsequent MRCPs did not identify any cholelithiasis, strictures or dilatations.

Eight years after choledochal cyst excision, the patient presented to the emergency department (ED) with epigastric and back pain, pale stools, raised inflammatory markers, epigastric tenderness and cholestatic liver function test (LFT) derangement and subsequently diagnosed with acute cholangitis. MRCP demonstrated several calculi in the intrahepatic biliary ducts with moderate left hepatic biliary duct ectasia (Fig. 1).

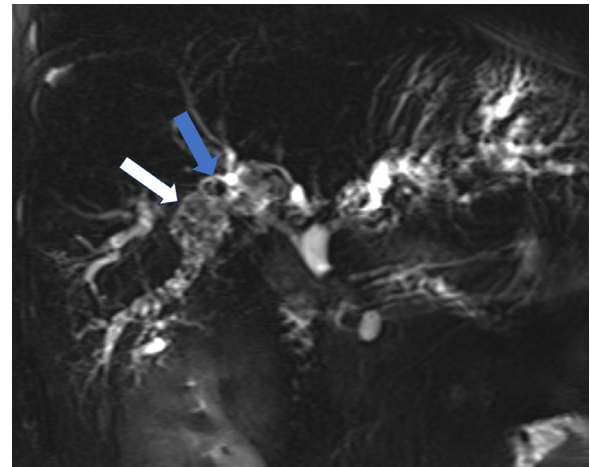


Fig. 1 – MRCP 8 years after choledochal cyst excision – Coronal T2 Half Fourier Single-shot Turbo-spin-Echo (HASTE) fat suppressed (FS) view of the biliary tree. This MRCP demonstrates several filling defects noted in moderately dilated intrahepatic bile ducts, largest measuring 8mm, with mild-to-moderate distal bile duct dilatation. White arrow: Dilated right posterior hepatic duct with multiple filling defects. Blue arrow: Large filling defect.



Fig. 2 – Initial PTC - demonstrating numerous intrahepatic filling defects in the right hepatic duct. White arrow: dilated right posterior hepatic duct. Blue arrow: filling defect.

The patient was commenced on intravenous antibiotics leading to clinical improvement. In the setting of clinical improvement, altered anatomy and an anticipated technically difficult procedure, ERCP was not pursued in the acute setting. The patient was subsequently discharged and elective PTC was arranged.

Two months later, the patient presented for PTC and biliary drain insertion. The right biliary duct was accessed percutaneously via segments 5/6 under ultrasound guidance. PTC demonstrated innumerable filling defects within the dilated biliary ducts (Fig. 2). A wire was passed beyond the biliary cal-

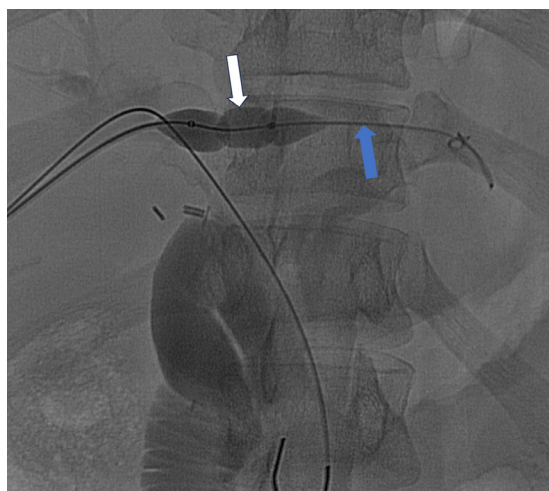


Fig. 3 – Repeat PTC - Image shows balloon dilatation of the left hepatic duct strictures. White arrow: Balloon dilatation at the site of a left hepatic duct stricture. Blue arrow: Catheter in the left hepatic duct.

culi through the hepaticojejunostomy and then into the jejunum. Balloon dilatation was performed on the biliary strictures at the level of the hepatic hilum. Stones were then trawled from the biliary tree into the jejunum and an external drain was left in situ. They were admitted post operatively for intravenous antibiotics and commenced on ursodeoxycholic acid to reduce stone formation.

Two weeks later, the patient presented for a second PTC and biliary drain insertion. Access to the left biliary tree was attempted percutaneously but due to technical difficulty and safety the left hepatic duct was accessed via the percutaneous right hepatic duct access. The PTC again demonstrated multiple intrahepatic stones. Stones from both the left and right hepatic ducts were trawled into the jejunum with a balloon. Multiple intrahepatic biliary strictures were dilated with a 20mm balloon (Fig. 3). Both left and right hepatic drains were left in situ.

Following this procedure, the patient developed hypotension and hypoxia secondary to multi-organism septicemia (*E coli*, *Klebsiella* and *Citrobacter*). Intravenous antibiotics were escalated leading to clinical improvement.

A progress MRCP was performed 1 month later. This demonstrated a partial reduction in biliary dilation, partial resolution of intrahepatic biliary calculi, and areas of biliary irregularity and stricture (Fig. 4).

Based on the MRCP findings and the unclear cause for the persisting pathology the multidisciplinary team concluded that drain exchange, drain upsize and percutaneous cholangioscopy would be appropriate management options. At PTC 1 month later, the smaller left sided drain was removed and cholangiogram demonstrated an irregular intrahepatic biliary tree with areas of dilation and filling defects in keeping with biliary stones. The right sided drain was exchanged and up-sized to a 20F drain.

The patient presented to ED 5 days postprocedure with right upper quadrant (RUQ) pain (presumed drain related) and

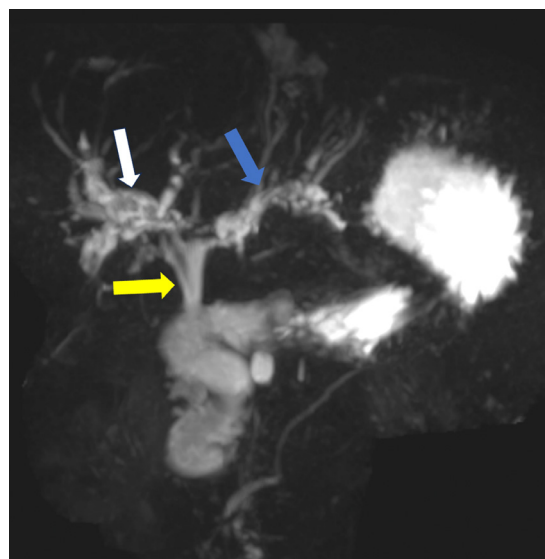


Fig. 4 – Progress MRCP 1 month after PTC - Coronal oblique maximal intensity projection (MIP) view demonstrates a reduction in biliary dilation and biliary calculi. However, there are areas of persisting ductal irregularity and stricture. Reduced dilation and internal filling defects seen in the right (white arrow) and left (blue arrow) hepatic ducts compared to the prior MRCP. Patent hepaticojejunal anastomosis (yellow arrow).

was subsequently discharged. Two weeks later, they represented to ED with RUQ pain, pale stools and deranged LFTs secondary to a dislodged right hepatic duct drain. Intravenous antibiotics were recommenced and the patient returned to the interventional radiology (IR) suite for migrated drain removal, re-establishment of the pre-existing percutaneous tract to the right duct, and 20Fr drain insertion.

During this admission, the first of 3 combined IR and gastroenterological procedures was performed (Fig. 5). The existing drain was removed, then using a stiff guide wire, an ultrathin endoscope was advanced into the right biliary tree via the established percutaneous tract. Stones were removed by Spy-glass basket (Boston Scientific, Marlborough, Massachusetts, USA) irrigation, and mobilization past the hepaticojejunal anastomosis. A balloon catheter was advanced to the hepaticojejunal anastomosis and inflated to 15 mm. Cholangiography was performed by injecting contrast via the ultrathin endoscope (Fig. 5). The biliary ducts were dilated although the left hepatic system was sub-optimally visualized despite multiple attempts. Deep biliary cannulation with a percutaneous catheter was also unsuccessful. A 20Fr percutaneous biliary drain was inserted into the right hepatic duct. Contrast was injected and adequate drain position was confirmed. The patient was discharged on oral antibiotics and planned for a repeat percutaneous cholangioscopy.

Due to recurrent cholangitis requiring intravenous antibiotics, the second combined IR and gastroenterology procedure was performed 1 month later. The existing right sided 20Fr biliary drain was removed and a wire was guided into the left hepatic ducts from the existing percutaneous tract. Percuta-

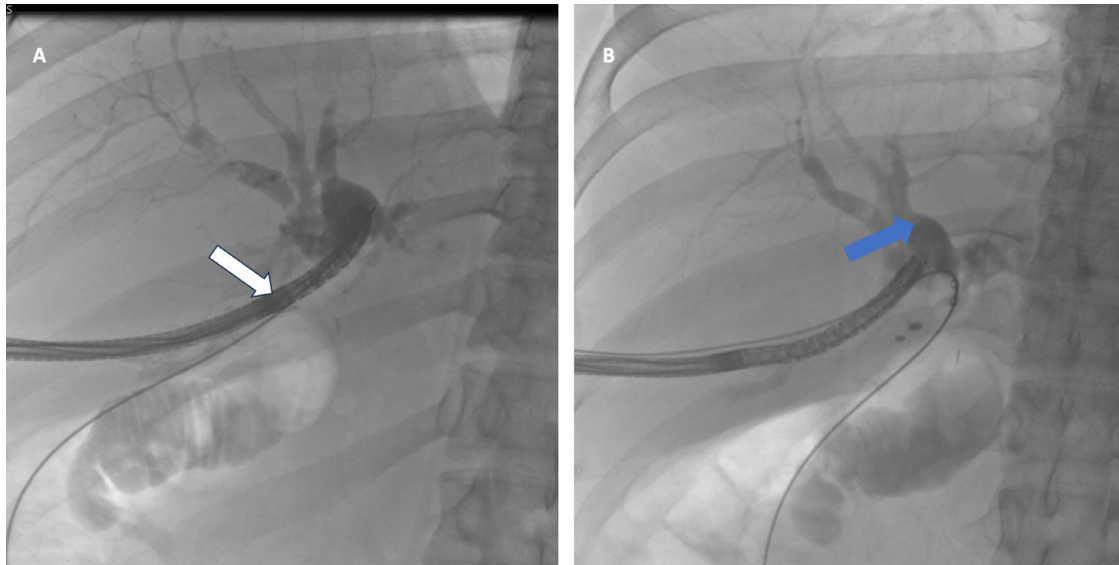


Fig. 5 – Endoscopic cholangiography - (A) Cholangiography of the right hepatic duct via an ultrathin endoscope at the start of the procedure. (B) Cholangiography of the right hepatic duct after biliary calculi removal and balloon plasty to the hepaticojejunal anastomosis. Image B shows a reduction in the number of biliary calculi seen as filling defects compared to the initial MRCP. White arrow: Ultrathin endoscope in the right hepatic duct. Blue arrow: Less dilated hepatic ducts with reduction in the number of filling defects.

neous cholangiogram demonstrated multiple left sided filling defects and duct dilatation. A SpyScope endoscope (Boston Scientific, Marlborough, Massachusetts, USA) was then inserted into the left intrahepatic ducts. Stricture and small stones were noted. The SpyScope endoscope was exchanged for a balloon biliary dilator, and the left hepatic duct was dilated to 12mm. Debris and stone fragments were then removed from the left hepatic duct to the jejunum using balloon trawl. The left hepatic duct was then visualized with an ultrathin endoscope but this was limited by debris and small clots. A drain was then placed into the left hepatic duct (Fig. 6) and another 20Fr drain was placed across the hepaticojejunos-tomy. The patient was covered with appropriate antibiotics post procedure.

The third and final combined IR and gastroenterological procedure was performed 3 weeks later. The two existing biliary drains were removed, then a wire was advanced to the hepaticojejunal anastomosis via the existing percutaneous biliary tract. An ultrathin endoscope was again inserted percutaneously via this same tract to assess the biliary anatomy. A few residual stones were noted in the left hepatic duct, these were mostly extracted with a Spyglass basket. The previously dilated left hepatic duct remained patent. The subsequent cholangiogram demonstrated left and right main hepatic duct patency with contrast draining well through the hepaticojejunal anastomosis, no filling defects were seen (Fig. 7). The endoscope was removed and the percutaneous tract was embolised with GelFoam (Pfizer, New York, New York, USA).

Following these procedures, the patient has been well. An MRCP was performed after 4 months, which demonstrated some ongoing intrahepatic biliary tree irregularity. There was reduced intrahepatic biliary dilatation overall and the number of filling defects had substantially decreased (Fig. 8). The

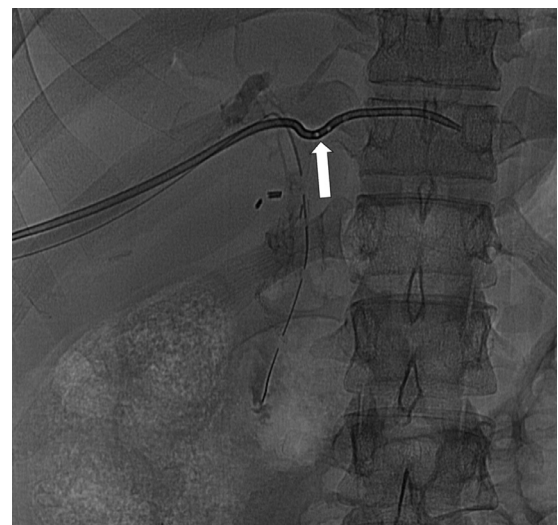


Fig. 6 – Spyglass cholangioscopy. (A) Cholangiogram post procedure demonstrating drain insertion to the left hepatic duct after biliary duct balloon dilatation. White arrow: Surgical drain sitting in the left hepatic duct.

most recent MRCP at 2 year follow up had an overall similar appearance with some mild persisting biliary dilatation with features of chronic inflammation (Fig. 9).

Discussion

Complications such as recurrent cholangitis, hepatolithiasis and anastomotic strictures are common after hepaticoje-

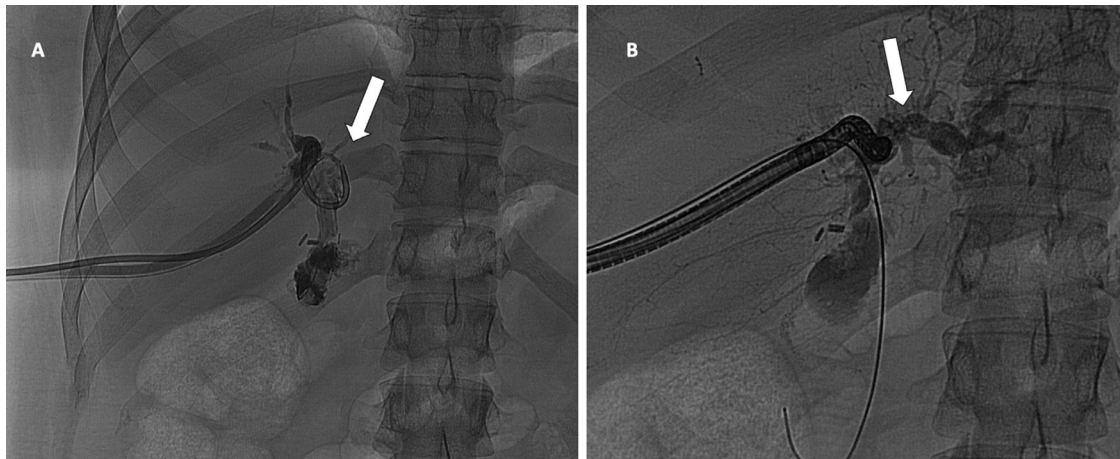


Fig. 7 – Endoscopic cholangiography - (A) Cholangiogram via pre-existing biliary drain demonstrating minimal contrast in the left hepatic duct (white arrow). (B) Cholangiogram post endoscopic SpyBasket removal of left hepatic duct calculi demonstrates increased patency of the left hepatic ducts (white arrow).

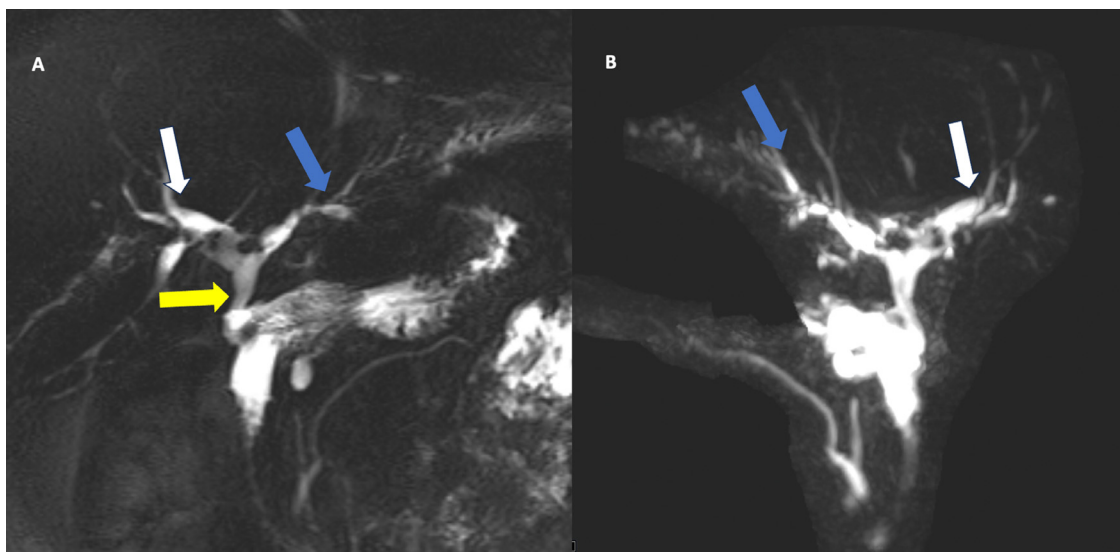


Fig. 8 – MRCP at 4 month follow up - Coronal T2 HASTE FS views (A + B) demonstrate significant reduction of biliary dilatation and calculi compared to the MRCP performed 1 year prior (Fig. 1). Reduced dilatation and internal filling defects seen in the right (white arrow) and left (blue arrow) hepatic ducts compared to the previous MRCP. Patent hepaticojejunal anastomosis (yellow arrow).

junostomy [5,6,11]. Recurrent cholangitis is associated with higher risk of strictures and is associated with male, sex, benign condition and post operative complications [5]. The underlying pathology is thought to be related to reflux of intestinal contents and pancreatic enzymes into the biliary tree and a longer roux limb for the hepaticojejunostomy may not prevent this reflux [12,13].

Biliary duct stones in patients with surgically altered anatomy are usually managed with ERCP but in patients with Roux-en-Y hepaticojejunostomy this can be technically difficult requiring double balloon enteroscopy, EUS-guided inter-

vention or a percutaneous approach [14–18]. This case highlights a minimally invasive, multidisciplinary approach to managing complications after hepaticojejunostomy in a patient with a previous biliary cyst.

In 2020, Tripathi et al presented 5 cases of percutaneous transhepatic cholangioscopy with the Spyglass endoscope for diagnostic and therapeutic management of biliary duct stones and strictures [19], 3 of which were patients with prior Roux-en-Y hepaticojejunostomy.

Kimura et al have described a case of cholangitis due to multiple intrahepatic ducts stones in a liver transplant pa-

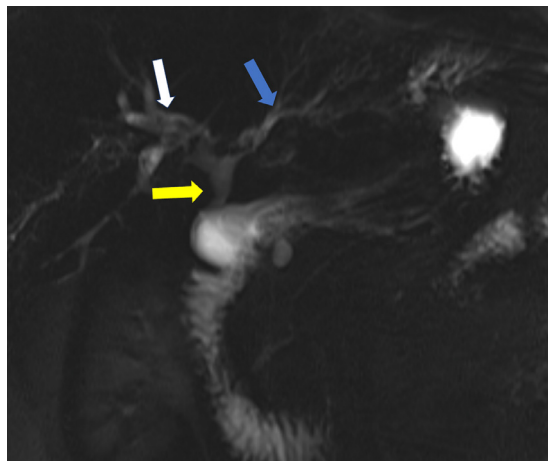


Fig. 9 – MRCP at 2 year follow up (Coronal T2 HASTE view) - Compared with the MRCP performed at 4 months the intrahepatic biliary tree dilatation is stable. Unchanged dilation and internal filling defects seen in the right (white arrow) and left (blue arrow) hepatic ducts compared to the prior MRCP. Patent hepaticojejunal anastomosis (yellow arrow).

tient with hepaticojejunostomy. The stones were removed with cholangioscopic lithotripsy and rendezvous technique using double balloon endoscopy [20].

To the authors knowledge this is the first case reported for the management of biliary calculi and strictures after Roux-en-Y hepaticojejunostomy for treatment of choledochal cysts and is the first published Australian case of a combined interventional radiology percutaneous approach and gastroenterological percutaneous endoscopic approach to management of biliary calculi and strictures.

Conclusion

This article outlines a minimally invasive, multidisciplinary approach to the management of intrahepatic biliary calculi and stricture after Roux-en-Y hepaticojejunostomy where traditional ERCP intervention is too technically difficult. This approach also allows for concurrent percutaneous approach with cholangiography and drain insertion with the ability to directly visualize the biliary tract and adds to the growing body of literature for the use of percutaneous transhepatic cholangioscopy.

Patient consent

The patient provided written consent for the publication of this report and ethics approval was received from the institutional Ethics Committee.

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