



Case Report

Percutaneous Transhepatic Cholangioscopy Utilizing Different Endoscopes to Optimize Treatment Success in Biliary Cast Syndrome, how a Paediatric Bronchoscope can Help: A Case Report

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Abstract

Biliary cast syndrome (BCS) is an uncommon complication after orthotopic liver transplantation (OLT), which can lead to serious clinical complications. Endoscopic removal of the biliary casts has gained popularity over the last years, especially following the latest technical advances in the field of cholangioscopy. Case report: We report a patient post liver transplant with hepaticojejunostomy (HJS) who underwent successful endoscopic removal of difficult to treat biliary cast via percutaneous access utilizing different endoscopes in a single session. Conclusion: Percutaneous single-operator cholangioscopy (pSOC) is a feasible and very useful method for diagnosing and treating patients with biliary cast syndrome, but in complex cases it requires high skill levels and different technical materials.

Keywords: Biliary cast syndrome; Cholangitis; Hepaticojejunostomy; Orthotopic liver transplantation; Percutaneous single-operator cholangioscopy; PTCD

Abbreviations: BCS: Biliary Cast Syndrome; ERCP: Endoscopic Retrograde Cholangio Pancreatography; HJS: Hepatico Jejunostomy; MRCP: Magnetic Resonance Cholangio Pancreatography; OLT: Orthotopic Liver Transplantation; pSOC: Percutaneous Single-operator Cholangioscopy; PTCD: Percutaneous Transhepatic Cholangiography and Drainage

Introduction

BCS is a relatively uncommon but severe complication after OLT, affecting 4-18 % of all OLT recipients. It is characterized by biliary obstruction caused by the presence of biliary casts and debris. Biliary cast formation is most frequently associated with biliary strictures following recurring infection or as a result of altered arterial flow of the liver [1]. Approximately 20 % of the affected patients require repeat-OLT [2]. In cases where a HJS has been performed, ERCP is not feasible and thus a percutaneous approach is usually the preferred alternative over surgery. Treatment can be very challenging and in order to enable cast removal various endoscopic techniques have already been [3-10] described including balloon/basket extraction with prior dilatation

of biliary strictures if necessary, electrohydraulic shock-wave or laser lithotripsy. Most patients who fail endoscopic and surgical treatment usually require re-transplantation.

Case

A 61-year-old patient underwent OLT due to (non)-alcoholic steatohepatitis (NASH/ASH)-associated liver cirrhosis and hepatocellular carcinoma. Three months postoperatively a liver biopsy was performed due to elevated liver function tests, which revealed intracellular damage due to either a biliary obstruction or drug induced. A postoperative endoscopic retrograde cholangiopancreatography (ERCP) showed no signs of biliary or anastomotic stenosis, so a tacrolimus induced or MMF-induced drug-induced liver injury (DILI) was postulated. After a switch of the immunosuppressive therapy to cyclosporine, he recovered only temporarily. Despite rapidly deteriorating liver function tests and renal failure resulting in a non-manageable clinical deterioration as reflected in his high lab MELD of 38, the patient underwent repeat-OLT. Explant histology showed unexpected signs of severe early chronic rejection. Two weeks later, he developed a purulent cholangitis. An ERCP revealed pus in the bile duct system and a small leakage of the choledochocholedochal anastomosis without significant stenosis or casts. Bile culture revealed a multidrug-resistant *Pseudomonas aeruginosa* strain. Despite biliary stenting

and antibiotic treatment, the patient did not recover and had to undergo surgery with resection of the extrahepatic bile ducts and conversion to a HJS due to severe ischaemic cholangiopathy. Four weeks postoperatively he once again developed cholestasis. A MRCP showed a dilation of the remaining sub hilar bile duct up to 10 mm with presence of filling defects as well as dilated intrahepatic ducts mainly in the left liver lobe without an obvious stenosis. Since an ERCP was deemed challenging due to the altered intestinal anatomy, a percutaneous transhepatic cholangiodrainage (PTCD) of the left liver lobe was performed. Cholangiography showed a dilation of the left intrahepatic bile ducts with filling defects in the remnant common bile duct. The HJS was dilated with a balloon up to four mm and an eight Fr Yamakawa drainage catheter was inserted. Bile cultures acquired during PTCD-insertion revealed once again a multidrug-resistant *Pseudomonas aeruginosa* strain. Two weeks later the patient was referred due to fever and cholestasis. A percutaneous cholangiography was repeated, which showed a persistent dilatation of the left intrahepatic bile ducts and filling defects proximal to the HJS. Following a dilatation of the percutaneous access up to 14 Fr and placement of a 12 Fr percutaneous sheath, a SpyGlass™ DS Discover cholangioscope (Boston Scientific) was inserted which revealed multiple biliary casts in the last 3 cm proximal of the HJS. Attempts to mobilize the casts with balloon catheters, electrohydraulic lithotripsy (EHL), basket and biopsy forceps were only partly successful, since some of the casts were strongly adherent to the biliary walls. The cholangioscope was exchanged to a Baby-Bronchoscope (Olympus EVIS EXERA III, BF-XP190). Following an intensive vigorous purging, the cast material could successfully be pushed through the HJS to the Jejunum (Figures 1-3). Consequently, a wire cannulation of the segment II was attempted, but due to the steep angle and a stenosis of the orifice, a fluoroscopic cannulation was not possible. Therefore, SpyGlass™ DS Discover was reinserted. Under direct cholangioscopic imaging a 0.025 in. guiding wire could be inserted to the left liver lobe (Figures 4-6). Subsequently a balloon dilatation of the orificium up to four mm enabled the complete scoping of the dilated left liver segment. Here no further stenosis or cast material were detectable, supporting the evidence that the previously dilated stenosis was the cause of the cholestasis in this segment. Finally, a 12 Fr Yamakawa drainage was placed successfully. Four weeks after the intervention the patient was stable with improved cholestatic liver function tests and the drainage was removed.

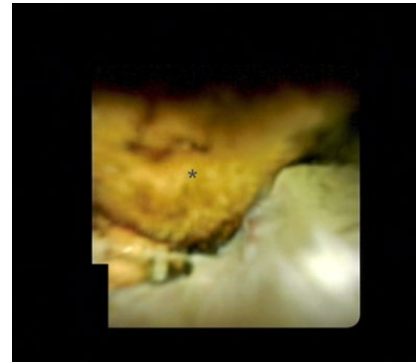


Figure 1: Cholangioscopic view via Spy DS Discover on cast material (asterisk) proximal to HJS.



Figure 2: Fluoroscopic view of cast material (asterisk) proximal to the HJS (arrow).

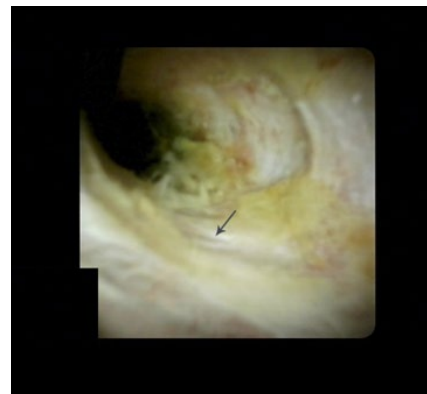


Figure 3: orifice of the liver segment II (arrow).

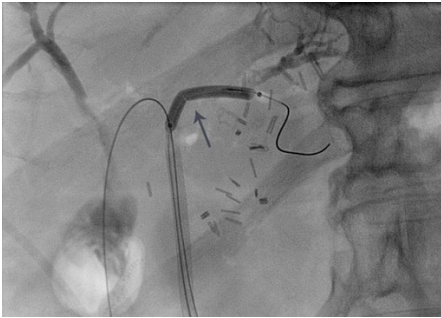


Figure 4: Dilatation of liver Segment II with a 4mm balloon.

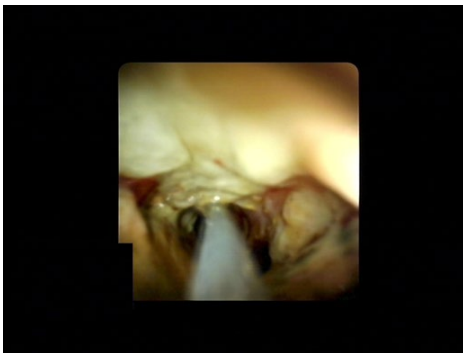


Figure 5: Hepatic duct proximal to the HJS with ischaemic type appearance after complete clearance of casts.

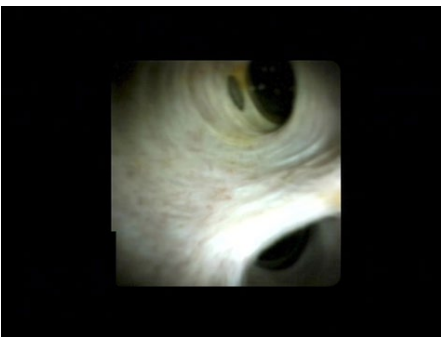


Figure 6: Normal appearing intrahepatic bile ducts in liver segment II.

Discussion

BCS, characterized by the presence of bile casts and debris, is usually a difficult to treat complication and is associated with a high morbidity, frequently requiring re-transplantation [2]. Over 70% of casts are identified in the first 4 months after OLT, but they can occur even several years after OLT. The most common underlying pathology is considered to be an ischemic injury of the biliary epithelium as is seen more frequently after transplantation of organs retrieved from donors after circulatory death [11], but other mechanisms such as anastomotic or non-anastomotic strictures

which lead to an accumulation of biliary sludge and formation of casts have also been proposed. Bacterial infection and biliary stones seem to be predisposing factors as well [12], in our case bile cultures revealed twice the presence of a multidrug-resistant *Pseudomonas aeruginosa* strain. Microcirculatory dysfunction appears to also play a role in the pathogenesis of casts. Bile ducts are fully dependent on arterial blood supply and hence prone for ischemia in case of hypoperfusion and microcirculatory failure [13], which can also explain the high prevalence of renal failure and need for renal replacement therapy in patients with BCS [14].

BCS can lead to cholangitis and in case of an intrahepatic biliary system involvement, management becomes more challenging. If a complete removal of casts is not feasible, there is a higher risk for the development of graft failure and need for re-transplantation [15]. In patients with typical signs such as fever, jaundice and elevated liver function tests, initial imaging modalities such as abdominal ultrasound and magnetic resonance cholangiopancreatography (MRCP) are strongly recommended. Since MRCP can have a varying sensitivity in diagnosing BCS in these critically ill patients (about 70%-90%) [15], a cholangioscopy assisted ERC is indicated in patients with normal biliary anatomy for further diagnostic evaluation and therapy.

Due to altered anatomy here pSOC was performed. The use of a SpyGlass Discover Digital Catheter (Boston Scientific, Natick, MA, USA) with a working length of 65 cm, especially designed for use via percutaneous access, provided direct visualization and prompt recognition of the obstructive cast material [16]. Furthermore, its 1.2 mm working channel allowed the insertion of several accessories such as biopsy forceps, an electrohydraulic lithotripsy probe as well as an extraction basket, in order to try to remove the cast material. It should be noted, that due to the left sided approach, the cholangioscope had to be bent in a U-shaped manner, which probably reduced its otherwise excellent manoeuvrability. This resulted in suboptimal clearance of the wall adherent cast material. For that reason, we opted for a 3.8mm paediatric bronchoscope (BF-3C160; Olympus Optical, Tokyo, Japan), the rigidity of which enabled us to mechanically scrape off the cast material and advance it through the HJS into the intestine.

Percutaneous cholangioscopy-guided stricture dilatation has advantages compared to cholangiography-guided intervention. The ischaemic biliary duct stenosis can sometimes be very narrow and almost impossible to cannulate. In our patient the cannulation of the dilated segment II was primarily not possible due to the steep angle and the pinhole-like stenosis of the orifice. Here, the SpyGlass Discover was once more used to aid this very challenging task. Thanks to its 4-axis steering capabilities, it was possible to advance a 0.025 in. guiding wire in the aforementioned segment. Balloon dilatation of the stenotic segment was performed which in turn allowed for direct visualisation of the Segment II. To

our surprise, there were no pathologic findings to be seen, which changed the management plan of this patient, since a left hemihepatectomy was also considered at that time (given the left sided bile duct dilatation).

Conclusion

The use of pSOC with direct visualisation of the biliary casts utilising a Spy-Discover-cholangioscope as well as a paediatric bronchoscope in the same setting did not only facilitate the diagnosis of BCS but was also crucial in clearing the intrahepatic casts as well as providing therapy changing diagnostic information in liver segments with difficult access due to stenosis. Particularly using a paediatric bronchoscope appears to provide add-on value for cases with adherent casts.

Disclosure Statement

The authors declare that there is no conflict of interest.

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