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Case Report

Intrahepatic lithiasis managed with hepatectomy and anterograde cholangioscopy through the right posterior sectorial duct

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ABSTRACT

Intrahepatic lithiasis, endemic in East Asia, is increasing in Western countries and without proper treatment, may cause cholangitis, liver abscesses, biliary cirrhosis, and cholangiocarcinoma. Management may require endoscopic, percutaneous, or surgical approach to the bile duct. Anterograde approach through the stump of the biliary duct during hepatectomy has been reported only rarely. We present a case managed through an anterograde approach, which is a valid alternative that preserves the integrity of the main biliary tree. A 76-year-old woman presented with recurrent severe cholangitis episodes and intrahepatic lithiasis in the right posterior sectorial bile duct, with a dominant stone in the junction with the left duct. Due to a variation in the biliary tree anatomy, the retrograde access was difficult and after undergoing several unsuccessful attempts at endoscopic clearance, she was offered surgical therapy. Right posterior sectionectomy and common bile duct anterograde exploration through the stump were performed, for stones clearance. The anterograde approach allowed complete hepaticolithothomy, preserving the main biliary tree and preventing anastomotic stricture or leakage. This technique should be considered in sectorial limited intrahepatic lithiasis with steep angle of drainage, unapproachable by retrograde access.

Keywords: Anterograde lithotomy, Intrahepatic lithiasis, Intraoperative cholangioscopy

INTRODUCTION

Hepatolithiasis is defined as the presence of calculi in the bile ducts peripheral to the confluence of the right and left hepatic ducts, regardless of the presence of lithiasis of the common bile duct (CBD) or gallbladder.[1] It is a frequent condition in East Asia where the relative incidence is 20-30% among all patients with gallstone disease, compared to 1% in the western countries. Asian studies report on larger series, and different etiologies have been observed. However, location of stones and technical options for treatment seem to not differ between the East and the West.[2]

No consensus has been reached on the ideal treatment for intrahepatic lithiasis. Several approaches have been proposed, including surgical and minimally invasive treatments like endoscopic retrograde cholangiopancreatography (ERCP) or percutaneous transhepatic cholangioscopy (PTCS). Still, stones impacted in peripheral ducts may not be amenable to choledocoscopic clearance, and residual and recurrent stones occur in 20% of patients treated with non-surgical strategies.[3] Furthermore, when biliary stenosis or hepatic atrophy is associated, hepatectomy has proven to provide lower recurrence rates with acceptable mortality risk, compared to non-surgical approaches and is therefore the treatment of choice. The strong association between intrahepatic cholangiocarcinoma and long-standing hepatolithiasis also reinforces the role of surgical treatment.[4] Moreover, hepatic resection is performed with declining morbidity and mortality rates, reflecting the increased expertise of hepatopancreatobiliary surgeons and improved perioperative management.[5]

It is known that patients with hepatolithiasis usually have concomitant extrahepatic duct stones, and biliary tract exploration is commonly required. On that account, during surgical intervention, choledochoscopy is typically performed via choledochotomy, to enable clearance of the main bile duct and ensure the absence of residual stones. Therefore, hepatectomy or segmentectomy are frequently associated with extraction of calculi through choledochotomy, with placement of a T-tube.

However, some concerns have been raised by those traditional approaches: choledochotomy often result in surgical complications like bile leak, biliary tract infection or T-tube displacement and discomfort.[6] Primary closure of the CBD has been presented as a safe alternative to T-tube placement, but this technique is not recommended in patients with un-dilatated CBD because of the stricture risk.[7] Furthermore, to address the CBD in a patient with history of previous biliary surgery may be challenging due to postoperative adhesions, responsible for a higher risk of iatrogenic injuries.[8] Biliary anastomoses have been performed for treatment of choledocholithiasis but are becoming controversial procedures, as high recurrence rates have been verified. Moreover, after bilioenteric anastomosis with a Roux-en-Y limb, endoscopic is precluded or at the very least extremely difficult to achieve.

An alternative bile duct exploration procedure during hepatectomy for hepatolithiasis have been pointed out only by a few authors, but showed encouraging results: to approach the distal orifice of the bile duct stump in the hepatectomy surface section as an entry path for exploration and removal of stones of the CBD and contralateral bile duct, with anterograde cholangioscopy performed through the same entry site.[8,9] This method avoids unnecessary choledochotomy and related risks. This approach, used in laparoscopic treatment of left-sided hepatolithiasis after left hepatectomy, showed significantly accelerated postoperative recovery compared with conventional choledochotomy, and satisfactory short- and long-term results.[10,11] The surgical approach may even be more relevant in right-sided lithiasis given the anatomical characteristics of the right biliary ducts. In particular, the right posterior sectorial bile duct (RPSBD) typically makes an abrupt angle when draining into the right anterior, the convergence or the left hepatic duct, known as Hjortsjö's crook.[12] This may impede retrograde approaches, mandating surgery.

We hereby illustrate a rare case of western non endemic hepatolithiasis of the RPSBD, associated with extrahepatic lithiasis, that was managed through hepatectomy, anterograde biliary cholangioscopy and hepaticolithotomy.

CASE REPORT

We report the case of a 76-year-old woman, with a history of open cholecystectomy 10 years earlier. In the past 5 years she presented with several episodes of cholangitis (grade II – Tokyo guidelines), with the need for hospital admission and intravenous antibiotics. Presenting symptoms included abdominal pain, nausea, vomiting, jaundice and fever.

Laboratory investigation revealed normal aspartate aminotransferase and alanine aminotransferase, normal or slight increase of bilirubin (1.7 mg/dl, normal 0.2-1.1 mg/dl), elevated gamma-glutamyl transpeptidase 144 U/l (normal up to 50 U/l) and alkaline phosphatase 257 U/l (normal up to 120 U/l). Abdominal ultrasound revealed a dilated intra and extrahepatic ducts. A magnetic resonance cholangiopancreatography (MRCP) described dilated CBD and right intrahepatic ducts, and lithiasis of the RPSBD (Figure 1). The dominant stone was located in the convergence of the RPSBD with the left hepatic duct, with a very abrupt Hjortjo's crook. A computed tomography (CT) angiography was performed and excluded iatrogenic vascular injury, namely to right hepatic artery. Endoscopic retrograde cholangiopancreatography with sphincterotomy was performed, resolving the choledocolithiasis, but was unable to reach the intrahepatic stone and didn't prevent new episodes of cholangithis.

Hepatolithiasis of the posterior right bile duct and the CBD persisted, causing renewed episodes of cholangitis during the following three years, after which the case was discussed at a multidisciplinary group meeting and right posterior sectionectomy was proposed.

During laparotomy, and using intraoperative ultrasound, the right posterior portal pedicle was isolated through an intrahepatic, extra-glissonian approach, for proximal control (Figure 2). The glissonian sheath was opened and the right posterior branches of the hepatic artery and portal vein were ligated, and the sectorial duct opened. Using a fiber cholangioscope, the right posterior duct and CBD were explored. Lithiasis of the RPSBD and a bigger stone in the confluence with the left hepatic duct were identified. Lithotripsy was achieved using shockwave, Mirizzi and Desjardins gallstone forceps and a Fogarty balloon-tipped catheter, in a retrograde direction (Figure 3).

To confirm successful clearance of the biliary tree, cholangiography was performed through the stump of the RPSBD. The orifice was then closed with a running monofilament polypropylene suture. Right posterior sector resection was then completed, with the liver parenchyma transected with the assistance of Cavitron ultrasonic surgical aspirator (CUSA), irrigated coupled bipolar cautery and polymer hem-o-lok ligating clips. No hepatic pedicle clamping was performed and the patient did not require intraoperative blood component therapy.

At day eight post-surgery, the patient presented one single episode of acute cholangitis, manifested with rigors and hypotension which responded with fluids and antibiotics (Dindo-Clavien grade II). She was discharged in the fifteenth day post-surgery. Pathology of the resected

specimen showed biliary lithiasis with chronic cholestasis and periportal fibrosis. Four years after surgery she is well, asymptomatic and with normal liver function tests. During the follow-up period, no hepatic lithiasis recurrence was detected and the patient did not experience further episodes of cholangitis.

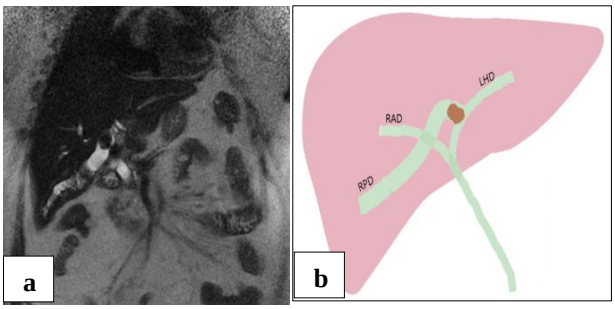

Figure 1: (a) MRCP describing dilated RPD (full arrow) and dominant stone located in the convergence with the LHD (doted arrow), and (b) schematic representation of the biliary anatomy, the dilated RPD and the location of the stone in the abrupt Hjortjo's crook.

RPD: Right posterior duct; RAD: right anterior duct; LHD: left hepatic duct

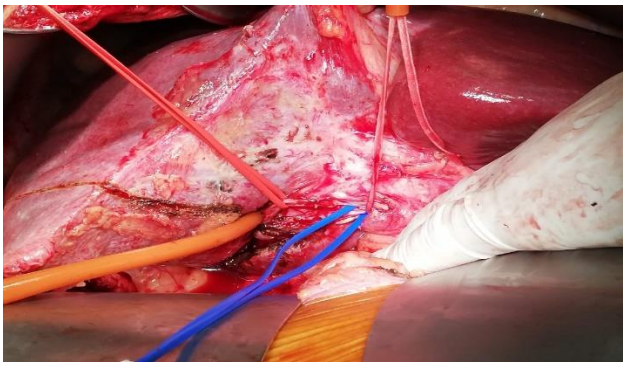

Figure 2: Dissection of the right posterior portal pedicle, with dissection of the right posterior branches of the hepatic artery and portal vein.

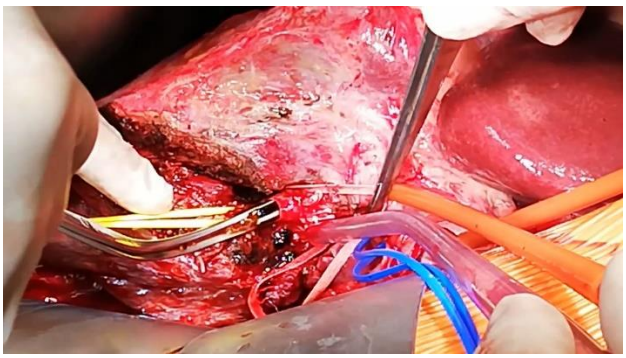

Figure 3: Right posterior sectorial duct opened before ligation and sectionectomy completion, for intraoperative biliary exploration and lithotripsy using gallstone forceps.

DISCUSSION

Hepatolithiasis is a condition that often leads to recurrent cholangitis and parenchymal atrophy, and in 60 to 73% of patients, concomitant extrahepatic stones are also present.[10] Therefore, aggressive treatment, associated with bile duct exploration with stone extraction are usually required. The patient we present had a history of multiple episodes of cholangitis and recurrence of intra and extrahepatic lithiasis after failure of endoscopic treatment attempts.

Predictors for failure of endoscopic CBD clearance such as previous biliary tract surgery, cholangitis, anatomic abnormalities and size of stones were potentially implicated.[7] PTCS is also a minimally invasive promising option and is used to treat intrahepatic stones by expanding the PTCS access gradually.[13] However, risk of bleeding, biliary fistula or infection is potentially increased in patients with a compromised biliary tract by previous infections.[14] In these cases, hepatectomy shows the most favourable outcome, as it removes all causative lesions with lower recurrence rate, also reducing the risk of developing cholangiocarcinoma.[15]

If extrahepatic stones are present at the moment of the parenchyma resection, biliary duct exploration is required. Conventional choledochotomy with cholangioscopy has been widely used for this purpose.[16] However, T-tube related complications cannot be underestimated, and are more evident in patients with previous biliary surgeries due to complex CBD dissection, as in the clinical case we present.[6]

Antegrade cholangioscopy and CBD exploration through the bile duct stump of hepatectomy cross section have advantages confirmed by several authors. Hwang et al demonstrated that this procedure avoided choledochotomy in 90.9% of patient and resulted in no residual stones and very low recurrence rate.[9] Since then, other studies continued to corroborate his findings and showed a reduced need for T-tube insertion; a shortened operative duration and in-hospital stay and decreased procedural complications associated with choledocholithotomy and CBD dissection, like biliary leak.[10,17,18]

In terms of recurrence, antegrade cholangioscopy has the advantage of following the direction of bile flow, lowering the chance of flushing out small gallstones into second or third order bile ducts, potentially causing residual stones. Hwang et al showed intrahepatic stone recurrence in three patients (6%), with a mean follow-up of 32 months, and another study showed recurrence in seven patients (18.9%), with a mean follow-up of 68 months – both with no differences between recurrence in the CBD group.[19] Regarding other common complications, one study identified three cases of iatrogenic injuries (duodenum, CBD and portal vein) in the group with CBD exploration, while such complications were avoided in the left hepatic duct (LHD) group.[18] Additionally, cholangioscopy via the

LHD stump was comparable to conventional choledochotomy in identifying secondary intra- or extrahepatic gallstones or biliary stenosis.[10]

The case we report has some particularities that are worth mentioning. First, it is a case of non-oriental primary intrahepatic lithiasis, a rare and sparsely published condition in the western world. The aetiology of primary intrahepatic lithiasis is yet to be fully understood, although four main factors are thought to be directly involved: cholestasis, infection, anatomic variants of the bile duct tree, and bile metabolic defects.[20] Variants of the ABCB4 and ABCB11 genes are associated with cholestasis and intrahepatic stones by influencing the content of phosphatidylcholine in bile and decreasing the secretion of bile acid, respectively.[21] Taking into consideration the patient's age, no testing was performed, but a possible genetic aetiology should be considered.

Secondly, this is a case of a right sided hepatolithiasis. The left hepatobiliary system is the most commonly involved, and approximately 81% of hepatolithiasis occurs in the left side.[6] This fact can be explained by the acute angle formed by the confluence of the LHD and the common hepatic duct, likely to cause cholestasis. Hepatectomy is rarely performed for unilateral right sided hepatolithiasis, and when bilateral lithiasis is present, left hepatectomy is preferred.[22] In a cohort study with 52 patients, the authors found a higher frequency of right hepatolithiasis (34,6%) when compared with previous studies. Some hypotheses were considered to explain this difference, such as the presence of anatomic variations in which the posterior segmental artery comes from the common hepatic artery or is divided and a branch runs through the gallbladder fossa, predisposing for injuries during cholecystectomy and causing ischemic cholestasis.[23] However, in the case we report no arterial injury was present. In addition, the development of right hepatolithiasis and abnormalities of the hepatic duct confluence were studied by Balandraud et al in a series of western patients and found that cranial shifts (causing acute angulation between the right and left hepatic ducts) of either the right lateral or the right paramedian sectorial duct were significantly correlated with right sided hepatolithiasis.[24] According to Couinaud's description in the general population, this type of variation occurs in 9.3%, and was found in 42,8% of patients in this study. In the case we report the patient present with an abrupt angle of the RPSBD draining into the left hepatic duct, as described by Balandraud et al.[25] We postulate that this contributed to the development of intrahepatic lithiasis of RSPD. Moreover, in the case we report this variation impaired retrograde access through ERCP.

However, the main original feature of the case herein described is the anterograde approach through the stump of B6/7, that allowed for the complete clearance of all stones, without significantly soling the remaining bile ducts with stone fragments. Even though the patient experienced one early episode of cholangitis, she remains otherwise well more than twelve months after surgery.

To our knowledge, this is the first case of a surgical anterograde approach for hepatolithiasis of the right lobe. Right-sided resection is a more complex surgical procedure, and right hepatectomy was found to be predictive of postoperative complications in patients with hepatolithiasis.[26] Thus, this method can simplify surgical exploration, minimizing the risks.

CONCLUSION

This new strategy applied for exploring and removing stones from the biliary tree during surgical resection is promising. The authors recommend that it should be considered in good surgical candidates with single-sector limited intrahepatic lithiasis, particularly with steep angle of drainage of right sectorial ducts, making retrograde approach less effective. However, more studies are needed specially to ascertain the value of this technique in particular patient populations, such as in patients with genetic cholestatic syndromes, who present a high recurrence rate.

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