TRANSPLANT INTERNATIONAL

Transplant International ISSN 0934-0874

LETTER TO THE EDITORS

Salvage of liver transplant with hepatolithiasis by percutaneous transhepatic cholangioscopic hepatolithotomy

doi:10.1111/tri.12387

Dear Sirs,

Hepatolithiasis, whilst endemic in East Asia, is rare in the West, where prevalence is less than 1% of all cholelithiasis [1]. The aetiology is incompletely understood, but biliary stasis, infection and excessive mucin production appear to be important factors [1,2]. Hepatolithiasis is associated with significant morbidity, often characterized by recurrent cholangitis that can result in the development of life-threatening sepsis, hepatic abscesses, secondary biliary cirrhosis and cholangiocarcinoma [1]. The management of hepatolithiasis is challenging, with high rates of treatment failure and recurrence. Although the standard approach to managing this condition is hepatic resection [2,3], percutaneous transhepatic cholangioscopic hepatolithotomy (PTCHL) offers a safe and minimally invasive alternative treatment option [1,4–7].

Little has been reported on the incidence or management of hepatolithiasis after liver transplantation. Here, we describe how PTCHL allowed us to salvage the graft and avoid the risks of abdominal surgery in a complex liver transplant recipient.

A 23-year-old female was referred to our centre for assessment and management of hepatolithiasis. She had undergone liver transplantation aged three in 1992 (segments 2 and 3 of an adult liver), for alpha-1 antitrypsin deficiency. Eleven days postoperatively, she required biliary reconstruction and had a Roux-en-Y hepatico-jejunostomy fashioned. Six months later, she presented with internal herniation through the roux-loop window in the transverse mesocolon resulting in extensive small bowel ischaemia. She had a stormy postoperative course, and over the subsequent 2 months, she underwent 10 relook laparotomies for various complications including further ischaemia and overall had approximately 170 cm of small bowel resected, including segments from the roux loop. (The hepatico-jejunostomy did not require revision.) She developed short bowel syndrome and remained dependent on total parenteral nutrition for many years. However, over last 5 years, she has managed to sustain with oral intake with periodic intravenous vitamin and mineral supplementation.

Although her liver graft function remained good throughout this period, she had intermittently deranged LFTs. A biopsy performed in 2010 had revealed a healthy graft. During the 6 months prior to referral, she experienced recurrent episodes of cholangitis with multiple hospital admissions. MRCP performed at the referring hospital demonstrated intrahepatic duct dilatation and the presence of intrahepatic filling-defects suggestive of stones which were confirmed on PTC. She was subsequently referred to our centre.

To clarify the ductal anatomy and assess the distribution of stones, we performed a further PTC. This confirmed multiple stones centrally within the intrahepatic ducts. Although the hepatico-jejunostomy was patent, the adjoining 5-cm segment of the roux loop was noted to be narrowed, but with free flow of contrast (Fig. 1a). CT angiography demonstrated patent hepatic arterial and portal venous inflow suggesting this was not a biliary cast syndrome (BCS).

A range of management options were considered by the multidisciplinary team, including liver resection, biliary exploration and retransplantation (potentially multivisceral in view of her short gut syndrome). Considering her history, prior good graft function and histology, and in the absence of intrahepatic cholangiopathy or biliary strictures, attempting to salvage her graft with minimally invasive PTCHL was felt to be most appropriate in the first instance. Our unit had previous experience of performing this procedure in the nontransplant setting.

The procedure was performed in conjunction with an interventional radiologist and urologist (with experience of percutaneous nephrolithotomy). Firstly, percutaneous transhepatic biliary drainage was established at an optimal position that would allow access to most of the biliary tree. This tract was allowed to mature for 3 weeks. Under general anaesthesia, the tract was dilated using serial dilators to permit introduction of a 28F Amplatz™ access sheath (Boston Scientific Corp Natick, MA, US). Using a nephroscope (26 Ch, Karl Storz), the intrahepat-

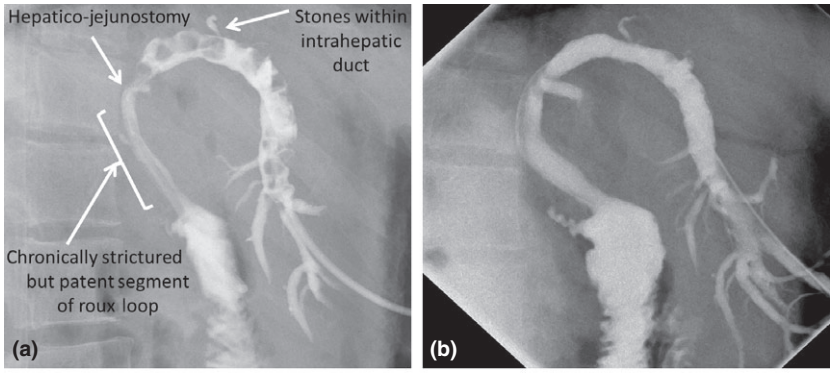

Figure 1 Percutaneous transhepatic cholangiogram images (a) prehepatolithotomy and (b) posthepatolithotomy. Stones within the intrahepatic ducts are clearly visible and have been successfully cleared.

ic stones were visualized. They were then broken down with an ultrasonic lithotripter with integrated suction (Swiss lithoclast Master, EMS), using the principles of percutaneous nephrolithotomy. The hepatico-jejunostomy and proximal roux loop were traversed using the flexible ureteroscope (DUR-8, ACMI), demonstrating them to be patent. Further branches of the intrahepatic ductal system were inspected using the flexible ureteroscope, and any stones seen in these branches basketed out using a ureteroscopic basket (Zero-tip, 1.9FR, Boston Scientific Corp.). Any remaining debris was extracted via the suction, or flushed distally. After clearance, the access sheath was removed and a large biliary drain sited. The patient made a good postoperative recovery and was discharged 4 days later. Her drain was gradually downsized and finally removed after 2 weeks. Completion cholangiogram, and follow-up MRCP, confirmed stone clearance, absence of intrahepatic ductal strictures and free flow through the roux loop (Fig. 1b). It has now been 20 months since the procedure, and the patient has experienced no further episode of cholangitis, her liver function tests have normalized, and she is now off long-term antibiotics.

Much of the experience with PTCHL comes from the Far East where this technique is established as a safe, minimally invasive treatment option for the clearance of intrahepatic calculi without sacrificing parenchyma [1–6]. It allows for direct visualization of the calculi and permits dilatation of any biliary strictures. Complete calculi clearance is achieved in 64–92% of cases which is comparable to surgical management [1,2,4]. Reported complications are predominantly minor despite the procedure tending to be reserved for poor surgical candidates. Major complications described include liver laceration, intra-abdominal abscess and disruption of the PTC tract, leading to bile leak and bleeding [1]. In the longer term, calculi and cholangitis recurrence is common, with rates ranging 20–63%, predominantly occurring in patients with incomplete calculi clearance and/or residual biliary strictures [2]. The advantage of PTCHL from this regard is that it offers the opportunity for repeated procedures without sacrificing liver parenchyma.

In liver transplant recipients with hepatolithiasis, in the absence of cholangiopathy and biliary strictures, as demonstrated by this case, PTCHL presents a minimally invasive and safe opportunity to salvage the graft.

Funding

Supported in part by the NIHR Cambridge Biomedical Research Centre. JA is supported by a Clinical Research Training Fellowship from The Wellcome Trust and a Raymond and Beverly Sackler Scholarship.

Jason M. Ali,[1] Teik C. See,[2] Oliver Wiseman,[3] William J. H. Griffiths[4] and Asif Jah[1]
1 Cambridge Transplant Unit, Cambridge University Hospitals NHS Trust, Cambridge, UK
2 Department of Interventional Radiology, Cambridge University Hospitals NHS Trust, Cambridge, UK
3 Department of Urology, Cambridge University Hospitals NHS Trust, Cambridge, UK
4 Department of Hepatology, Cambridge University Hospitals NHS Trust, Cambridge, UK
e-mail: asif.jah@addenbrookes.nhs.uk

References

1. Mori T, Sugiyama M, Atomi Y. Gallstone disease: management of intrahepatic stones. *Best Pract Res Clin Gastroenterol* 2006; **20**: 1117.
2. Cheon YK, Cho YD, Moon JH, Lee JS, Shim CS. Evaluation of long-term results and recurrent factors after operative and nonoperative treatment for hepatolithiasis. *Surgery* 2009; **146**: 843.
3. Ramia JM, Palomeque A, Muffak K, Villar J, Garrote D, Ferron JA. Indications and therapeutical options in hepatolithiasis. *Rev Esp Enferm Dig* 2006; **98**: 597.
4. Chen C, Huang M, Yang J, *et al.* Reappraisal of percutaneous transhepatic cholangioscopic lithotomy for primary hepatolithiasis. *Surg Endosc* 2005; **19**: 505.
5. Kow AW, Wang B, Wong D, *et al.* Using percutaneous transhepatic cholangioscopic lithotripsy for intrahepatic calculus in hostile abdomen. *Surgeon* 2011; **9**: 88.
6. Lee JH, Kim HW, Kang DH, *et al.* Usefulness of percutaneous transhepatic cholangioscopic lithotomy for removal of difficult common bile duct stones. *Clin Endos* 2013; **46**: 65.
7. Cheung MT, Wai SH, Kwok PC. Percutaneous transhepatic choledochoscopic removal of intrahepatic stones. *Brit J Surg* 2003; **90**: 1409.

