Retrospective analysis of surgical management options of hepatolithiasis in a tertiary care centre in Bangladesh

Ahmed AHMT^a, Hassan RM^a, Raihan HMS^b, Arefin MS^c, Islam MA^d, Rashid MM^e

ABSTRACT

Background: Hepatolithiasis (HL) is the presence of stones in the bile ducts proximal to the confluence of the hepatic ducts. It is endemic in the Asia-Pacific region. The etiology of hepatolithiasis is not fully understood but genetic, dietary and environmental factors are thought to be contributory. Malnutrition and low socioeconomic conditions are associated with a high incidence of intrahepatic stones. This study aimed to analyze the demographic and clinicopathological factors, appraise the role of surgical management and analyse the postoperative outcomes of HL in a tertiary care centre in Bangladesh.

Methods: This study was conducted in the Department of Hepato-Biliary-Pancreatic Surgery and Liver Transplant (HBPS & LT), BIRDEM General Hospital, Dhaka, Bangladesh. Retrospective data of all patients operated for HL from January 2011 to December 2022 were analyzed with regard to clinical parameters, biochemical parameters and different types of surgical management.

Results: A total of 52 patients underwent surgical treatment for HL. Of the patients, 65.4% were female and 34.6% were male. Of the patients, 94.2% had abdominal pain, 57.7% had fever and 28.8% presented with jaundice. A total of 30.8% of patients had a history of previous biliary surgery. Liver atrophy was present in 38.5% of cases. Hepaticojejunostomy (HJ) alone was done in 36 (69.2%) patients, hepatic resection with bilioenteric anastomosis was done in 10 (19.2%) patients and hepatic resection with T-tube placement in common bile duct (CBD) done in 6 (11.5%) patients. Endoluminal access loop was done in 2 (3.8%) patients. On follow-up visits, 60% of patients were asymptomatic. There was no postoperative mortality.

Conclusion: Results from present study revealed encouraging results with minimal post operative complications and no mortality.

Key words: hepatolithiasis, hepatic resection, hepaticojejunostomy, choledocholithiasis, bilioenteric anastomosis.

BIRDEM Med J 2024; 14(3): 137-142

DOI: https://doi.org/10.3329/birdem.v14i3.76238

Author information

- A.H.M. Tanvir Ahmed, Muhammad Rafiqul Hassan, Registrar, Departtment of Hepato-Biliary-Pancreatic Surgery and LiverTransplant (HBPS & LT), BIRDEM General Hospital, Dhaka, Bangladesh.
- b. Hossain Mohammad Sabbir Raihan, Assistant Professor, Department of Surgery, KhwajaYunus Ali Medical College Hospital, Sirajgonj, Bangladesh.
- c. Mirza Shamsul Arefin, Assistant Professor, Department of Surgery, BIHS General Hospital, Dhaka, Bangladesh.
- d. Md. Anisul Islam, Registrar, Department of Urology, Bangladesh Medical College Hospital. Dhaka, Bangladesh.
- e. Md.Mamunur Rashid, Professor and Head, Department of HBPS & LT, BIRDEM General Hospital, Dhaka, Bangladesh.

Address of correspondence: A.H.M. Tanvir Ahmed, Registrar, Department of HBPS & LT, BIRDEM General Hospital, Dhaka, Bangladesh. E-mail: drtanvir_ahmed@yahoo.com Received: February 24, 2023 Revision received: August 22, 2024

Accepted: August 27, 2024

INTRODUCTION

Hepatolithiasis (HL) is defined by stones present proximal to the junction of the right and left hepatic ducts causing complete or intermittent biliary obstruction resulting in recurrent attacks of abdominal pain, fever and jaundice (Charcot's triad).¹ HL is endemic in East Asia, where prevalence can range up to 50%.² Intrahepatic stones are uncommon in western countries with an incidence of about 1%.^{3,4} However, the incidence is steadily increasing owing to increasing immigration.⁵ HL typically develops between the ages of 30 and 70 years, with the highest incidence in the fifth and sixth decades of life.⁶ Incidence is equal in both male and female. It does, however, differ greatly by country, with differences even within subpopulations.¹ The cause of HL is unknown but ethnic factors, poor nutrition, bacterial infection, parasite infestation and bile duct anatomy anomalies have all been suggested. Chronic parasitic infestations, particularly *Clonorchis sinensis* as a result of raw fish consumption and *Ascaris lumbricoides* are more prevalent in South-East Asian countries and detected in up to 30% of patients with HL.⁷⁻⁹ HL is also known as "Oriental cholangiohepatitis" or "pyogenic cholangitis" and is associated with a high rate of treatment failure and recurrence.¹⁰

Dong et al. described a classification of HL for use in determining surgical approaches.¹¹ Type 1 is a localized disease and type 2 contains diffusely distributed multiple HL and is divided into three different subcategories. The presence of extra-hepatic stones in this classification is defined as type E with three subgroups.¹¹

Complications of HL include suppurative cholangitis, liver abscess and chronic disease leading to biliary cirrhosis and cholangiocarcinoma; early treatment may prevent additional liver damage and improve long-term prognosis.¹⁰ The goal of treatment to cure continuing infections, avoid recurring cholangitis and consequent hepatic fibrosis, minimize the need for repeated instrumentation and prevent the emergence of cholangiocarcinoma, regardless of the source.¹¹

According to several studies, the following are the indications for hepatectomy as a therapy for HL: (i) unilobar HL, (ii) atrophy of the affected liver segments or lobe, (iii) presence of a liver abscess, (iv) cholangiocarcinoma and (v) multiple intrahepatic stones causing marked proximal biliary stricture or dilation.¹²⁻¹⁶ Access to the biliary tree is critical in the treatment of HL to treat residual and recurring stones as well as ductal

strictures using endoscopic or percutaneous procedures. Stone retrieval through the T-tube tract or by an access loop implanted as a stoma¹⁷ or in the subparietal layer¹⁸ is addressed via puncture under fluoroscopic guidance. Alternatively, the Roux limb of the hepaticojejunostomy (HJ) can be anastomosed to the stomach¹⁹ or duodenum²⁰ for future endoscopic management. This study was designed to evaluate the demographic and clinicopathological factors, appraise the role of surgical management and analyse the postoperative outcomes of patients with HL.

METHODS

This retrospective study was conducted at BIRDEM General Hospital in Dhaka, Bangladesh. Selective patients with HL who were operated in this hospital during the period between January 2011 to December 2022 were included in this study. Data were analyzed from a maintained institutional database; 52 patients were analyzed in this study. Patients were evaluated clinically like abdominal pain, fever and jaundice. Previous investigations and treatment history including endoscopic and surgical interventions were reviewed. Complete blood count, liver function tests and renal function tests were done in all patients. Patients were evaluated initially by ultrasound (USG) and computed tomography (CT) (Figure 1 a,b). Magnetic resonance imaging (MRI) with magnetic resonance cholangiopancreatography (MRCP) (Figure 2) was done to determine the location of stones, strictures and the presence of lobar atrophy or liver abscess and treatment was planned accordingly. Based on the presence of stones and segmental liver atrophy, patients were classified into 1a (right HL), 1b (left HL), 1c (bilateral HL) and surgical management options were chosen accordingly. Postoperative complications were recorded.



Figure 1. Contrast-enhanced CT scan showing **a.** Choledocholithiasis with hepatolithiasis in left ductal system **b.** Bilateral hepatolithiasis.



Figure 2. MRCP revealing dilated CBD and left intrahepatic ducts with filling defects

RESULTS

Total patients were 52 with female predominance (34, 65.4%). Mean age was 42 years. Common symptoms were abdominal pain, fever and jaundice. Demographic and clinical characteristics are shown in Table I. Six (11.5%) patients had liver abscess. Twenty six (50%) patients had bilateral HL, 19 (36.5%) had left HL and 7 (13.5%) had right HL. Associated gallstones and common bile duct stones were seen in 34 (65.38%) patients. Liver atrophy was seen in 20 (38.5%) cases. Fifteen (28.8%) cases showed segment 2 and 3 atrophy and five (9.5%) cases

Table I. Demographic and clinical features of patients with hepaticolithiasis (N = 52)

| Parameter | N (%) |
|--------------------------|------------------|
| Age (range) | 42 years (15-67) |
| Sex | |
| Male | 18 (34.6) |
| Female | 34 (65.4) |
| Symptoms | |
| Abdominal pain | 49 (94.2) |
| Fever | 30(57.7) |
| Jaundice | 15 (28.8) |
| Charcot triad | 11 (21.2) |
| Location of calculi | |
| Right HL | 7(13.5) |
| Left HL | 19 (36.5) |
| Bilateral HL | 26(50) |
| Previous biliary surgery | 16 (30.8) |

HL: hepatolithiasis.

showed segment 2, 3 and 4 atrophy. Two cases had dead worm in intrahepatic duct (Figure 3a).



Figure 3. Operative pictures of resected left lobe of liver showing **a**. Hepatolithiasis with self knotted worm **b**. Hepatolithiasis with ductal stricture.

Sixteen (30.8%) patients had a history of previous surgery; cholecystectomy in 7 (13.5%) patients, cholecystectomy with common bile duct (CBD) exploration and T-tube drainage in 5 (9.6%) and cholecystectomy with choledochoduodenostomy (CDD) in 4 (7.7%) patients. During revision surgery Roux-en Y HJ was done for all these 16 patients after complete clearance of stones (Figure 4b). Patients with no prior biliary surgery (36) underwent hepaticolithotomy with Roux-en-Y HJ in 20 cases (38.5%). Hepatic resection was done in 16 patients; right hepatectomy in one, left hepatectomy in 4 and left lateral sectionectomy in 11 cases. In hepatic resection cases Roux-en-Y HJ was done in 10 (19.2%) cases and T-tube placement was done in 6 (11.5%) cases. Endoluminal gastric access loop was made for two (3.8%) HJ patients.



Figure 4. Operative pictures **a.** Resected left lobe of liver showing hepatolithiasis with choledocholithiasis **b.** Biliary enteric bypass (Roux-en-Y Hepaticojejunostomy).

Twenty (38.5%) patients had growth in from bile culture, 15 (28.8%) had *Klebsiella pneumoniae* and five (9.6%) had *Escherichia coli*. Five (9.6%) patients had operative wound infection which were managed by regular dressing and wound closed by secondary stiches. Pulmonary complications like pneumonitis developed in two (3.8%) patients. Postoperatively bile leak developed in one (1.9%) hepatic resection patient and was managed by per-cutenious ultrasono guided drainage. At median follow-up of 28 months (range: 2-36 months), 37 patients were symptom free, 45 (86.53%) patients had no recurrence, 7 (13.46%) had recurrence of HL; reoperation was done in three (5.76%) after 7 years. Other four was managed by ERCP. There were no 30 and 90 day mortality. Histopathological report of resected liver confirmed recurrent pyogenic cholangitis with an evidence of cirrhosis in 6 (11.5%) patients (Figure 5b) and one (1.9%) patient had cholangiocarcinoma.



Figure 5. Operative picture- Effect of Hepatolithiasis **a.** Multiple cholestatic abscess in left lobe of liver **b**. Micro and Macro nodular Chirrhotic change in left lobe of liver.

DISCUSSION

Hepatolithiasis is an uncommon illness in western countries and endemic in East Asian countries.¹¹ There is no clear consensus regarding the most effective treatment. In the present study, it was noted that 26 (50%) cases had bilateral HL and 19 (36.5%) had left HL. In Mohanraj et al.²¹ study, eight out of 14 patients had left HL and only two cases had bilateral HL. Chen et al.¹⁴ study showed that 77% of patients had stones confined to the left lobe of the liver and 14% had stones in both lobes of the liver.

In the present study, 49 (94.2%) patients had pain and only 30 (57.7%) had a fever, which is similar to most studies where almost all the patients presented with pain and fever with a mean duration of four years.^{10,13,21}

The major goals of treating HL are to remove all stones and eliminate bile stasis in the biliary system to prevent recurrence and infection. As a result, the management must be tailored to the disease's various presentations. Surgical treatment options include biliary decompression with bilio-enteric anastomosis with/ without hepatic resection. Over the last few decades, both the techniques of liver resection and perioperative management have significantly improved, resulting in a remarkable decline in the morbidity and mortality of hepatic surgery.¹⁴

Hepatectomy is the best approach for treating HL because it removes both stones and strictured bile duct and resects the atrophic portion of the liver, thus reducing the risk of recurrent stones and eliminating the potential presence of cholangiocarcinoma.¹³ Hepatic resection with HJ or T-tube was done in the present study in 16 (30.8%) cases. Hepatic resection was performed on those cases that showed evidence of liver atrophy, cholestetic abscess or secondary biliary chirrhosis on imaging. There was no in-hospital mortality and a low morbidity rate (15.38%). All three patients who had previously undergone HJ underwent anastomosis revision.

The stone recurrence rate after the complete stone removal was 13.46% in our series. Despite best efforts, none of the existing therapeutic approaches can guarantee total clearance and post-treatment follow-up is necessary to deal with residual or recurring stones. Recurrent stones and cholangitis occur with a frequency of 9.5-16% following surgery and 31-40% after

endoscopic lithotomy.²² Despite developments in different methods of treatment for hepatolithiasis, important challenges to success include intrahepatic biliary strictures, impacted calculi and unreachable peripheral calculi.²³ Advantages of doing access loop formation allow future endoscopic access to manage recurrent or residual stones without the need for surgery.^{17-20,24} Kassem et al.²⁵ reported successful treatment of remnant stones and recurrent stones in their series of 42 permanent access cases (hepaticocutaneous jejunostomy). To conclude, our result from a resource constrain setting is encouraging.

Authors' contribution: MIA and MMR planned study. ATMTA drafted manuscript. All authors read and approved final version for submission.

Conflicts of interest: All authors declare the following:

REFERENCES

- Lorio E, Patel P, Rosenkranz L, Patel S, Sayana H. Management of hepatolithiasis: review of the literature. Curr Gastroenterol Rep 2020, 22:30.
- Catena M, Aldrighetti L, Finazzi R, Arzu G, Arru M, Pulitanò C, et al. Treatment of non-endemic hepatolithiasis in a Western country. The role of hepatic resection. Ann R Coll Surg Engl 2006, 88:383-9.
- Shoda J, Tanaka N, Osuga T. Hepatolithiasis epidemiology and pathogenesis update. Front Biosci 2003, 8:e398-409.
- Pausawasdi A, Watanapa P. Hepatolithiasis: epidemiology and classification. Hepatogastroenterology 1997; 44: 314-6.
- 5. Li C, Wen T. Surgical management of hepatolithiasis: a minireview. Intractable Rare Dis Res 2017; 6:102-5.
- Freise J, Mena J, Wen KW, Stoller M, Ho S, Corvera C. A rare presentation of hepatolithiasis in an adolescent patient: a case report. Int J Surg Case Rep 2020; 72:343-5.
- Tazuma S. Gallstone disease: epidemiology, pathogenesis, and classification of biliary stones (common bile duct and intrahepatic). Best Pract Res Clin Gastroenterol 2006; 20:1075-83.
- Leung JW, Yu AS. Hepatolithiasis and biliary parasites. Baillieres Clin Gastroenterol 1997; 11:681-706.
- Kim HJ, Kim JS, Joo MK. Hepatolithiasis and intrahepatic cholangiocarcinoma: a review. World J Gastroenterol 2015; 21:13418-31.
- Huang MH, Chen CH, Yang JC. Long-term outcome of percutaneous transhepatic cholangioscopic lithotomy for hepatolithiasis. Am J Gastroenterol 2003; 98:2655-62.

- Feng X, Zheng S, Xia F, Ma K, Wang S, Bie P, et al. Classification and management of hepatolithiasis: a highvolume, single-center's experience. Intractable Rare Dis Res 2012; 1:151-6.
- Uchiyama K, Onishi H, Tani M, Kinoshita H, Ueno M, Yamaue H. Indication and procedure for treatment of hepatolithiasis. Arch Surg 2002, 137:149-53.
- Chijiiwa K, Yamashita H, Yoshida J, Kuroki S, Tanaka M. Current management and long-term prognosis of hepatolithiasis. Arch Surg 1995; 130:194-7.
- Chen DW, Tung-Ping Poon R, Liu CL, Fan ST, Wong J. Immediate and long-term outcomes of hepatectomy for hepatolithiasis. Surgery 2004; 135:386-93.
- Sakpal SV, Babel N, Chamberlain RS. Surgical management of hepatolithiasis. HPB (Oxford) 2009; 11:194-202.
- Tabrizian P, Jibara G, Shrager B, Schwartz ME, Roayaie S. Hepatic resection for primary hepatolithiasis: a singlecenter western experience. J Am Coll Surg 2012, 215: 622-6.
- Barker EM, Winkler M. Permanent-access hepati cojejunostomy. Br J Surg 1984; 71:188-91.
- Beckingham IJ, Krige JE, Beningfield SJ, Bornman PC, Terblanche J. Subparietal hepaticojejunal access loop for the long-term management of intrahepatic stones. Br J Surg 1998; 85:1360-3.
- Sitaram V, Perakath B, Chacko A, Ramakrishna BS, Kurian G, Khanduri P. Gastric access loop in hepaticojejunostomy. Br J Surg 1998; 85:110.

- Jameel ARA, Pitchaimuthu A, Raju P, Shanmugasundaram R, Obla NB, Gounder KD. Hepatico-jejuno-duodenal access loop - a modified biliary reconstruction technique for facilitated endoscopic access to biliary tree following surgery for hepatolithiasis. Int J Hepatobiliary Pancreat Dis 2018; 8:100080-4.
- Mohanraj K, Joeimon JL, Aravind A, Solomon R, Balamurali R, Ramkumar G, et al. Hepatolithiasis in south Indian scenario- a case series. IOSR J Dent Med Sci 2017; 16:53-6.
- 22. Otani K, Shimizu S, Chijiiwa K. Comparison of treatments for hepatolithiasis: hepatic resection versus cholangioscopic lithotomy. J Am Coll Surg 1999, 189:177-82.
- Wen XD, Wang T, Huang Z, Zhang HJ, Zhang BY, Tang LJ, et al. Step-by-step strategy in the management of residual hepatolithiasis using post-operative cholangioscopy. Therap Adv Gastroenterol 2017; 10:853-64.
- Hamad MA, El-Amin H. Bilio-entero-gastrostomy: prospective assessment of a modified biliary reconstruction with facilitated future endoscopic access. BMC Surg 2012; 12:9.
- Kassem MI, Sorour MA, Ghazal AH, El-Haddad HM, El-Riwini MT, El-Bahrawy HA. Management of intrahepatic stones: the role of subcutaneous hepaticojejunal access loop. A prospective cohort study. Int J Surg 2014, 12:886-92.