

Original Research Article

Treatment outcome of hepatolithiasis: Nepalese experience

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ABSTRACT

Background: Hepatolithiasis is a rare disease with high rate of treatment failure and recurrence. This study aims to review the burden, management and outcome from an endemic region.

Methods: A retrospective review of database of patients with hepatolithiasis managed surgically from 2015 to 2019 was performed. Diagnosis was based on the clinical findings and radiological investigations. Demographic data, clinical presentation, extent of disease and type of surgical management were evaluated. The outcome measures included immediate stone clearance, postoperative complications and follow-up.

Results: Hepatolithiasis was seen in nine (0.34%) out of 2,600 patients being evaluated for gallstone disease. Three patients were young, while the remaining six were in the middle-age group. The presenting symptoms were pain abdomen (78%) and jaundice (22%). Hepatolithiasis was located in the left, right and both ductal systems in 5, 1 and 3 patients respectively. Liver resection for unilateral disease was done in 3 patients: left hepatectomy (n=2) and left lateral segmentectomy (n=1). High bile duct exploration and bilio-enteric drainage was done in 5 patients. One patient required hepatolithotomy and T-tube drainage due to cholangitis. Complete stone clearance was achieved in 78%. Complications included surgical site infection and cholangitis in 2 patients. There was no operative mortality. Histopathology revealed recurrent pyogenic cholangitis. At median follow-up of 28 months, 7 patients are symptom-free.

Conclusions: Hepatectomy is an effective treatment when disease is confined to the left lobe. Combined surgical procedure is an acceptable option for bilateral or right-sided hepatolithiasis.

Keywords: Gallstones, Hepatolithiasis, Hepatectomy, Surgery

INTRODUCTION

Hepatolithiasis is the presence of stones proximal to the confluence of the left and right hepatic ducts, irrespective of the presence of gallstones and extra hepatic bile duct stone disease.¹ The disease is difficult to treat because of the high rate of treatment failure and recurrence. Long-standing hepatolithiasis can lead to recurrent pyogenic cholangitis, liver abscess, portal hypertension, secondary biliary cirrhosis and cholangiocarcinoma.^{2,3} The disease is more common in East Asia (Taiwan, Japan and Korea)

and is endemic in Asia-pacific region, with prevalence as high as 30-50%.¹ It occurs more commonly in 5th to 6th decades of life, with no gender predominance. Most of the patients present with non-specific abdominal pain but patients can be asymptomatic. Hepatic resection (best option), stone removal with bilio-enteric drainage alone or in combination, and percutaneous transhepatic cholangioscopic lithotomy are the primary treatment modalities.^{2,4} The aim of this study was to review the burden and surgical outcomes of patients with hepatolithiasis from an endemic region.

METHODS

A retrospective review of prospectively maintained database was performed. All patients treated for hepatolithiasis from June 2015 to July 2019, in the Division of Surgical Gastroenterology, Department of Surgery were included. Ethical approval to conduct the study was obtained from BPKIHS Institutional Review Committee.

The inclusion criteria were all patients requiring surgery for the hepatolithiasis. Those patients who refused for surgery (n=2) were excluded. The diagnosis of hepatolithiasis was made based on the clinical and radiological (ultrasound, computed tomography (CT) and MRI) investigations. Demographic data, preoperative biliary drainage, prior surgery, site of stones in the intrahepatic biliary system, associated cholelithiasis and choledocholithiasis were recorded for all patients. Need for liver resection and type of surgery performed were also included. Outcomes included immediate stone clearance rate, postoperative complications, death and short-term follow-up.

In presence of choledocholithiasis, bile duct exploration was done in common hepatic duct and extended to left duct (if required) for wide bilio-enteric anastomosis. The intraoperative choledochoscopy was performed to confirm stone clearance from the intrahepatic and extra hepatic ductal system. Statistical analysis was performed with SPSS v 17.0 software for the descriptive statistical analysis by calculating mean, median, standard deviation and percentage where appropriate.

RESULTS

Out of 2,600 patients being evaluated for gallstone disease and planned for cholecystectomy during the study period, 9 (0.34%) patients had hepatolithiasis. Three patients were young (16-23 years), while the rest were in middle age (40-52 years) group. There was female (n=7, 78%) predominance. The most common presentation was pain in the abdomen (78%), while the jaundice was seen in only 2 (22%) patients. Two (22%) patients required preoperative biliary drainage: one for concomitant distal bile duct stone with cholangitis requiring endoscopic biliary drainage, and the other right percutaneous transhepatic biliary drainage for cholangitis (Table 1).

The distributions of hepatolithiasis were as follows: left duct in 5 (56%) patients, bilateral in 3 (33%) and on the right duct in 1 (11%) patient. Two (22%) patients underwent prior cholecystectomy at another center. Associated gallstones and common bile duct stones were seen in 6 (66%) patients each. The atrophy-hypertrophy complex due to the recurrent cholangitis and stricture was seen in 2 patients, both on the left side. There was no evidence of portal hypertension or secondary biliary cirrhosis.

Table 1: Clinical profile and treatment outcome of patients with hepatolithiasis.

Parameters	N (%)
Age, mean (range)	37 (16-52)
Sex (male:female)	2:7
Presenting illness	
Pain abdomen	7 (78)
Jaundice	2 (22)
Cholangitis	2 (22)
Prior cholecystectomy	2 (22)
Preoperative biliary drainage	2 (22)
Site of lesion	
Left duct	5 (56)
Right duct	1 (11)
Bilateral	3 (33)
Associated cholelithiasis/choledocholithiasis	6 (66)
Atrophy-hypertrophy complex	2 (22)
Operative procedure	
Left hepatectomy	1 (11)
Left hepatectomy+HJ	1 (11)
Cholecystectomy,bile duct exploration, HJ	5 (56)
Cholecystectomy, T-tube drainage	1 (11)
Left lateral segmentectomy, HJ	1 (11)
Complete stone clearance	7(78)
Operative morbidity	2(22)
Follow-up, median (months)	28 (2-36)

Liver resection was performed in 3 (33%) patients (left hepatectomy 2; left lateral segmentectomy-1); two required concomitant hepaticojejunostomy. In the 5 (56%) patients, high common bile duct exploration, stone extraction and bilio-enteric drainage (Roux-en-Y hepaticojejunostomy) was done (Figure 1 and 2). The remaining one (11%) patient underwent cholecystectomy, left duct stone extraction (via common hepatic duct) and T-tube drainage (without bilio-enteric drainage) due to the presence of pus in the biliary tract suggestive of severe cholangitis. The indications of liver resections were presence of stones localized in a unilateral lobe and atrophy of the affected segments (Figure 3). Complete intrahepatic stone clearance was seen in 7 (78%) patients. Two (22%) patients had residual stones, both in the right secondary duct which could not be retrieved by intraoperative cholangioscopy. Postoperatively, two (22%) patients had wound related complications (n = 1) and cholangitis (n=1), which were managed conservatively. There were no 30- and 90- day mortality. Pathological report of resected liver confirmed recurrent pyogenic cholangitis, without any evidence of cirrhosis or malignancy. At median follow-up of 28 months (range: 2-36 months), 7 patients are symptom-free, while the remaining two patients with residual stones are receiving medical management (Table 1).

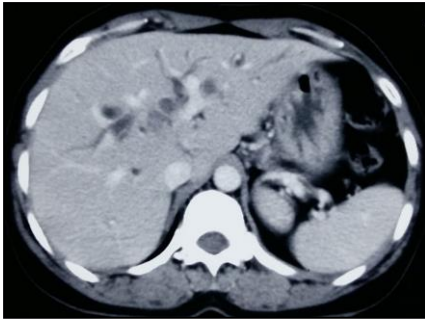


Figure 1: Contrast-enhanced CT scan showing bilateral hepatolithiasis.



Figure 2: Intraoperative view showing multiple (22 in number) blackish gallstones in the left, right and common hepatic ducts.

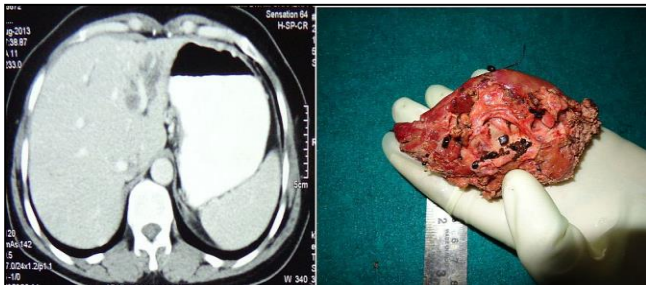


Figure 3: Isolated left hemi-liver hepatolithiasis with liver atrophy, requiring left hepatectomy.

DISCUSSION

In the present study, although our region falls under endemic zone for hepatolithiasis, the disease was detected in only 0.3% of cases with gallstone disease. However, population-based studies are required to determine the exact prevalence of hepatolithiasis in Nepal. The disease was predominantly seen in the young and middle-age population, although commonly described in the elderly population.⁵ This early-onset disease may be because of the malnutrition, low socioeconomic status and recurrent subclinical infections of the biliary tree.¹ Fortunately, none of the patients had secondary biliary cirrhosis or cholangiocarcinoma, which has been described in approximately 8% and 10% of patients respectively.^{3,6}

A previous study had reported the left lobe, right lobe and both lobes to be involved in 49%, 13% and 38% respectively, which is similar to that observed in this study.² If hepatolithiasis is confined to the left liver, liver resection (left hepatectomy or left lateral segmentectomy) is the best treatment.^{4,7} In the present study, it was performed in 33% of the patients. Hepatectomy is the most effective treatment as it not only removes all the stones, but also takes care of the strictures, atrophic segments, potentially carcinomatous bile ducts thus reducing the risk of recurrent hepatolithiasis.¹ If the hepatolithiasis is right-sided or bilobar, percutaneous transhepatic cholangioscopic lithotomy or lithotripsy is the initial treatment method of choice.^{3,8} If failed, it should be followed by hepatectomy of the severely affected segments, combined with stone removal, on-table choledochoscopic removal of stones from the remnant liver (with laser lithotripsy) and biliary bypass to prevent recurrence.^{2,9-11} Biliary bypass (Roux-en-Y hepatico-jejunostomy) facilitates post-operative natural clearance of residual intrahepatic stones. Bilobar disease is categorized as a high grade disease with greater risk of disease recurrence, often requiring multi-modality treatment for optimal management.¹² Surgery in bilobar disease is associated with higher morbidities and requires multi-segmental resection.

Surgical treatment remains an independent prognostic factor for a lower disease recurrence. In a recent study by Tan et al, from Singapore General Hospital, which included 157 patients (106 required surgical treatment) of hepatolithiasis, liver resection (with parenchymal disease) and biliary bypass (without parenchymal disease) was better than non-surgical treatment (ERCP and PTC) for disease recurrence.¹³ Parenchymal disease was defined as the presence of liver atrophy and/or abscess. In their cohort, 43.9% of patients experienced at least one episode of disease recurrence through their course of clinical follow-up. Any surgical intervention or common bile duct exploration with biliary bypass was associated with decreased risk of disease recurrence.

The stone clearance rate following treatment for hepatolithiasis (non-surgical and surgical) ranges from 60-83%, and is highest for hepatectomy.³ In the present study it was seen in 78%, which matches with the published study reports.² Similarly, recurrence is the most worrisome part of the hepatolithiasis after all the interventions, hence long-term follow-up is required. It occurs in 20% of patients after complete stone clearance, and is greater for patients having intrahepatic strictures. Similarly, after incomplete stone clearance, it occurs in 57% of patients.^{1,3}

CONCLUSION

In conclusion, hepatolithiasis, although rare, is becoming increasingly evident in young and middle age group Nepalese patients. Hepatectomy is an effective treatment when disease is confined to the left lobe. Combined

surgical procedure (stone extraction, on-table choledochoscopy and bilio-enteric drainage) is an acceptable option for bilateral or right-sided disease.

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Ethical approval: The study was approved by the Institutional Ethics Committee

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