

## Case Report

# Surgical management of hepatolithiasis: an institutional experience of a rare disease

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### ABSTRACT

Hepatolithiasis remains as a rare condition in the majority of western world countries. Although genetic, dietary and environmental factors have been associated with the disease, the exact etiology of this entity remains elusive. Several approaches; surgical and non-invasive have been well described. However, surgery remains the standard of care for definitive treatment of the disease. We sought to present the recent experience of our clinic regarding the surgical management of hepatolithiasis in the background of a narrative literature review. All adult patients with hepatolithiasis admitted to our center during the period 2018 to 2020 were included in the study. Patients' demographics, comorbidities and preoperative characteristics were collected by chart review. All patients underwent open left lateral hepatectomy. Three cases with hepatolithiasis were identified. All of them were male and had been suffering from recurrent pyogenic cholangitis. All patients remained asymptomatic and disease-free through the entire follow-up, except one case that was complicated with bile leakage during the immediate postoperative period. Hepatolithiasis is currently a rare condition in the western world. Surgery is safe and effective treatment approach for refractory cases and, provided that it is performed by experts in hepatobiliary surgery, it remains imperative for the definitive treatment of the disease.

**Keywords:** Hepatolithiasis, Surgery, Left lateral hepatectomy

### INTRODUCTION

Hepatolithiasis, although prevalent in East Asia, remains as an uncommon entity in most western countries.<sup>1</sup> However, immigration waves from countries with high prevalence are expected to change the epidemiological status of the disease in the western world.<sup>2,3</sup> Genetic, dietary and environmental factors are considered to be contributory to the disease.<sup>3,4</sup> However, the etiology of hepatolithiasis has yet to be fully determined. The goal of hepatolithiasis treatment is to resolve ongoing infections, prevent recurrent cholangitis and subsequent hepatic fibrosis, avoid multiple instrumentations and minimize the potential of progression to cholangiocarcinoma.<sup>5</sup>

Despite that conservative management options have been well described and established in the literature; surgery remains the standard of care for the definitive treatment of the disease. Particularly, sectional hepatectomy is considered as the best approach for the treatment of calculi of the intrahepatic ducts, since it removes stones, along with the strictured intrahepatic duct, resects the atrophic portion of the liver and, additionally, eliminates the potential of cholangiocarcinoma development.<sup>6,7</sup> In current study, we sought to present the recent experience regarding the surgical management of recurrent, symptomatic left hepatolithiasis in the background of a narrative literature review.

**CASE REPORT**

**Patients' characteristics**

Patients with confirmed diagnosis of hepatolithiasis who were admitted in our clinic from 2018 to 2020 were included in the study. Patients' demographics, comorbidities and preoperative characteristics were retrospectively collected via chart review. All patients underwent open left lateral hepatectomy for the treatment of recurrent left hepatolithiasis and were followed-up over a period of six months.

**Surgical approach**

A left lateral hepatectomy was the procedure performed in all cases of our series. Particularly, on the right side of the falciform ligament, a dissection of the left hepatic artery was performed. Then, the left portal vein was ligated close to the umbilical fissure. A transection of the liver parenchyma followed, one centimeter from the

falciform ligament, with concomitant ligation of the feedback vessels intraparenchymal. Finally, the left hepatic duct was ligated. On the left side of the falciform ligament, the left hepatic vein is transected with a 45 mm Echelon vascular stapler.

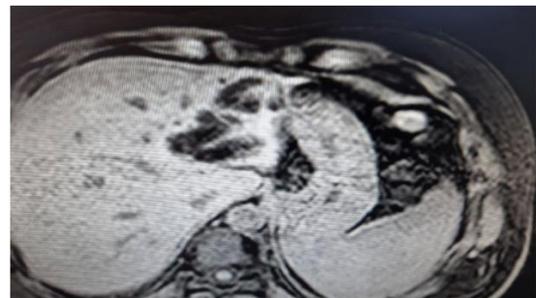
Over a three-year period, three cases were included in current report. The subjects to the procedure were all males, Caucasians, who had a history of recurrent episodes of acute pyogenic cholangitis, managed preoperatively with repeated endoscopic retrograde cholangiograms along with plastic stent placement. In our series, one case was complicated with bile leakage within 12 hours post-discharge that required readmission and reoperation. All patients remained asymptomatic and disease-free through the entire follow-up period. Patients' demographics, comorbidities and peri-operative characteristics are shown in (Table 1). Preoperative imaging characteristics of the disease are depicted in (Figures 1-4). Pictures from the operative room after the resection of the diseased liver segments are demonstrated in (Figures 5 and 6).

**Table 1: Characteristics of patients who underwent left lateral hepatectomy for isolated left-sided hepatolithiasis.**

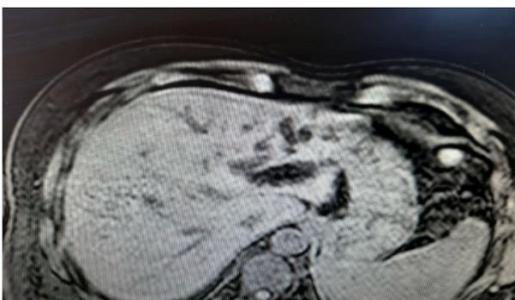
Case	Age	Gender	Race	CCI	Preoperative diagnosis	Number of preoperative ERCPs	Readmission	Complications
1	43	Male	White	0	Left lobe hepatolithiasis	9	Yes	Bile leakage
2	55	Male	White	1	Left lobe hepatolithiasis	7	No	No
3	53	Male	White	1	Left lobe hepatolithiasis	7	No	No



**Figure 1: Preoperative abdominal MRI of a patient with left-sided hepatolithiasis.**



**Figure 3: Preoperative abdominal MRI of a patient with left-sided hepatolithiasis.**



**Figure 2: Preoperative abdominal MRI of a patient with left-sided hepatolithiasis.**



**Figure 4: Preoperative abdominal MRCP of a patient with left-sided hepatolithiasis.**



**Figure 5: Intraoperative image after a left lateral hepatectomy.**



**Figure 6: Intraoperative image after a left lateral hepatectomy.**

## DISCUSSION

In light of the ongoing and upcoming advances in the field of non-surgical procedures for the treatment of hepatolithiasis, the role of surgery should be clearly determined. It has been stressed in the literature that surgery is an essential part of multidisciplinary management of recurrent pyogenic cholangitis with hepatolithiasis.<sup>8-10</sup> In current case report, the experience regarding a disease that, albeit prevalent in East is revealed, it remains frankly rare in the western world. Multiple studies from high volume tertiary centers have suggested that segmental hepatectomy is safe and effective and, therefore should be the treatment of choice for the definitive treatment of intrahepatic lithiasis. Nuzzo et al argued that partial hepatectomy allows for definitive treatment of primary intrahepatic lithiasis and prevention of cancer.<sup>11</sup> Furthermore, Catena et al suggested that surgery for single lobe hepatolithiasis should be indicated early after diagnosis, since this strategy may reduce the mortality and morbidity rates of hepatic resection.<sup>1</sup> Additionally, Uchiyama et al showed that hepatectomy appeared to be by far the most effective treatment for patients with isolated hepatolithiasis in terms of recurrence rate, compared to other modalities, such as percutaneous transhepatic cholangioscopic lithotripsy (PTCSL), cholangioenterostomy and T-tube drainage.<sup>12,13</sup> On the other hand, less invasive methods, namely shockwave and laser lithotripsy, are considered to be alternative procedures that offer a fair chance of

clearing the stones in patients who are not excellent candidates for surgery.<sup>14</sup> Therefore, non-surgical procedures such as cholangiography, although limited in their therapeutic capabilities, may play a vital role in the diagnosis and preoperative evaluation.<sup>15</sup>

## Limitations

Current study carries some limitations. Since it was a report of few cases, it included a small number of observations. Additionally, the patients who underwent hepatectomy were healthy individuals with very few comorbidities. Hence, by definition, it was not powerful enough to be generalizable in terms of mortality and complication rates. However, the approach of division in regard to the management of recurrent isolated hepatolithiasis was in accordance with the current evidence published in the literature. Moreover, there have not been previous studies about hepatolithiasis in our country, attesting to the fact that the disease has low prevalence in our country. Overall, this was a single-center study presenting experience in the surgical management of a rare and challenging disease.

## CONCLUSION

Hepatolithiasis is currently a rare condition in the western world but the dynamic of its epidemiology is yet to be revised in the future. Despite the rapid advances in minimally invasive procedures, surgery is safe and effective and, provided that it is performed by experts in hepatobiliary surgery, it remains imperative for the definitive treatment of the disease.

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