

## Surgical management of refractory hepatolithiasis

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### ABSTRACT

*Hepatolithiasis, or intrahepatic bile duct stones, is a condition characterized by the formation of calculi within the bile ducts of the liver. It is prevalent in Southeast Asia but rare in Western countries. Complications include recurrent biliary colic, jaundice, and cholangitis, potentially leading to liver abscesses and secondary biliary cirrhosis. This case report discusses a 42-year-old Asian man with a history of choledocholithiasis and hepatic cholelithiasis, presenting multiple times with obstructive jaundice and ascending cholangitis. Despite multiple ERCP procedures and sphincterotomies, CT imaging revealed persistent intrahepatic lithiasis confined to the right hepatic lobe. The patient underwent right hepatectomy and Roux-en-Y hepaticojejunostomy. His postoperative course was uneventful, with rapid recovery and successful follow-up. Hepatolithiasis management is challenging due to high recurrence rates and complex treatment requirements. Risk factors include bile stasis, infections, biliary anomalies, and dietary habits. This case underscores the importance of comprehensive surgical intervention, meticulous preoperative planning, and multidisciplinary care for managing refractory hepatolithiasis. Effective management of refractory hepatolithiasis involves advanced surgical approaches and long-term follow-up to prevent recurrence and improve outcomes. Future research should focus on novel therapeutic strategies to enhance patient care.*

**Keywords:** General surgery, hepatolithiasis, hepaticojejunostomy, refractory, biliary pathology

### INTRODUCTION

Hepatolithiasis, also known as intrahepatic bile duct stones, is a condition characterized by the formation of calculi within the bile ducts of the liver.<sup>1</sup> This condition is relatively rare in Western countries but is endemic in Southeast Asia, characterized by either complete or intermittent biliary obstruction proximal to the joining of the right and left hepatic ducts, causing recurrent attacks of abdominal pain, fever, and jaundice.<sup>2</sup> Hepatolithiasis can lead to significant health issues, including recurrent abdominal pain, liver abscesses, and secondary biliary cirrhosis.<sup>2</sup> The exact cause of hepatolithiasis remains unclear, but contributing factors

may include bile stasis, bacterial infection, and genetic predispositions.<sup>3</sup>

Diagnosis typically involves imaging techniques such as ultrasound or computed tomography (CT). These imaging modalities are essential for identifying the presence, size, and location of intrahepatic stones, as well as any associated biliary dilation or liver parenchymal changes. Additionally, laboratory tests often reveal elevated liver enzymes and bilirubin levels, indicative of biliary obstruction and liver inflammation.

Effective management is crucial to prevent complications of refractory hepatolithiasis and prevent future recurrences.<sup>4</sup> Treatment can range from endoscopic retrograde cholangiopancreatography (ERCP) for removal of the stones to more invasive surgical intervention in severe cases.<sup>5</sup> This case report discusses a patient with refractory hepatolithiasis, highlighting the challenges in diagnosis and management,

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**DOI:** 10.12746/swjm.v13i55.1459

**Table 1. Laboratory Values of Patient on Day of Admission to ER**

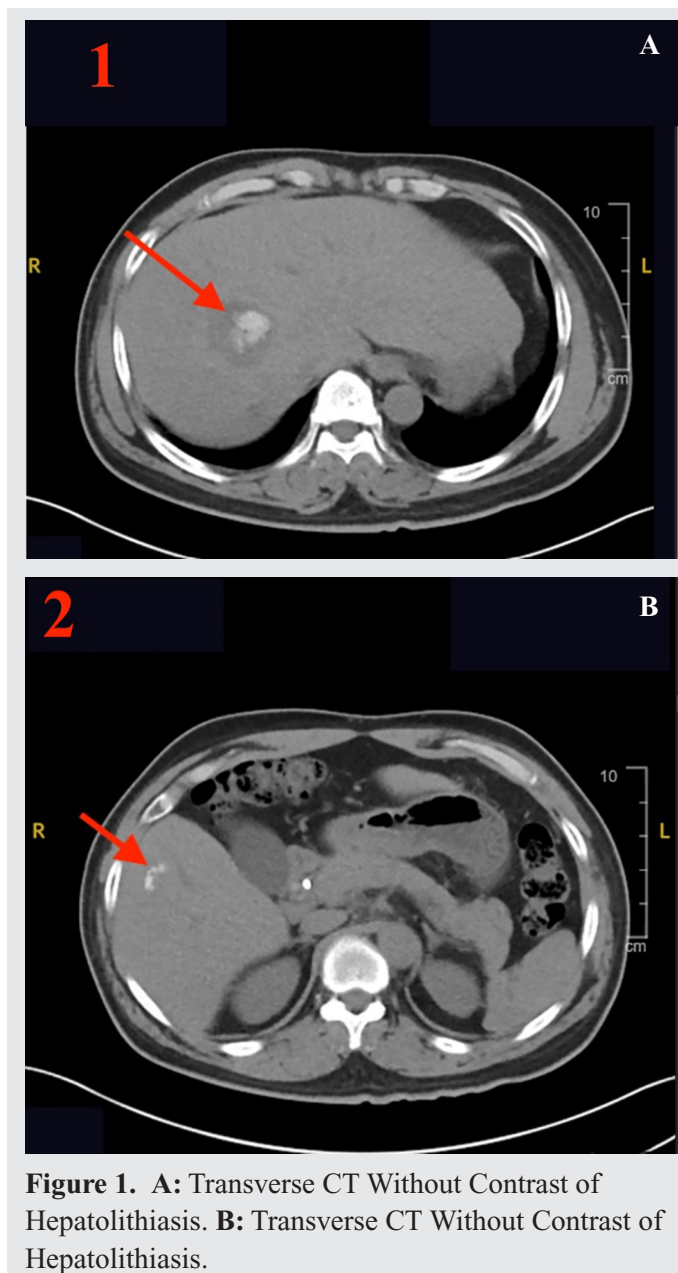
Laboratory Values	Patient Values	Reference Ranges
Hemoglobin (G/DL)	8.6	13.5–17.5
WBC ( $\times 10^3$ /MCL)	6.2	4,500–11,000
Platelets ( $\times 10^3$ /MCL)	138	150,000–400,000
ALT (U/L)	72	10–40
AST (U/L)	81	12–38
T. Bili (U/L)	1.51	0.1–1.0
Alk Phos (U/L)	37	25–100
Albumin	2.9	3.5–5.5
Creatinine (Mg/dL)	0.6	0.6–1.2

and providing insights into potential strategies for improving patient outcomes.

## CASE

A 42-year-old Asian man with a history of cholelithiasis and hepatic cholelithiasis was admitted multiple times with obstructive jaundice and ascending cholangitis (Table 1). The patient presented with colic right upper quadrant pain, associated with nausea and vomiting. On examination, he was found to have right upper quadrant tenderness. He had undergone multiple ERCP procedures involving stone extractions and two sphincterotomies to relieve his jaundice. Computed tomography imaging revealed intrahepatic lithiasis confined to the right hepatic lobe, with no involvement of the left lobe (Figures 1A, 1B, 2A, and 2B).

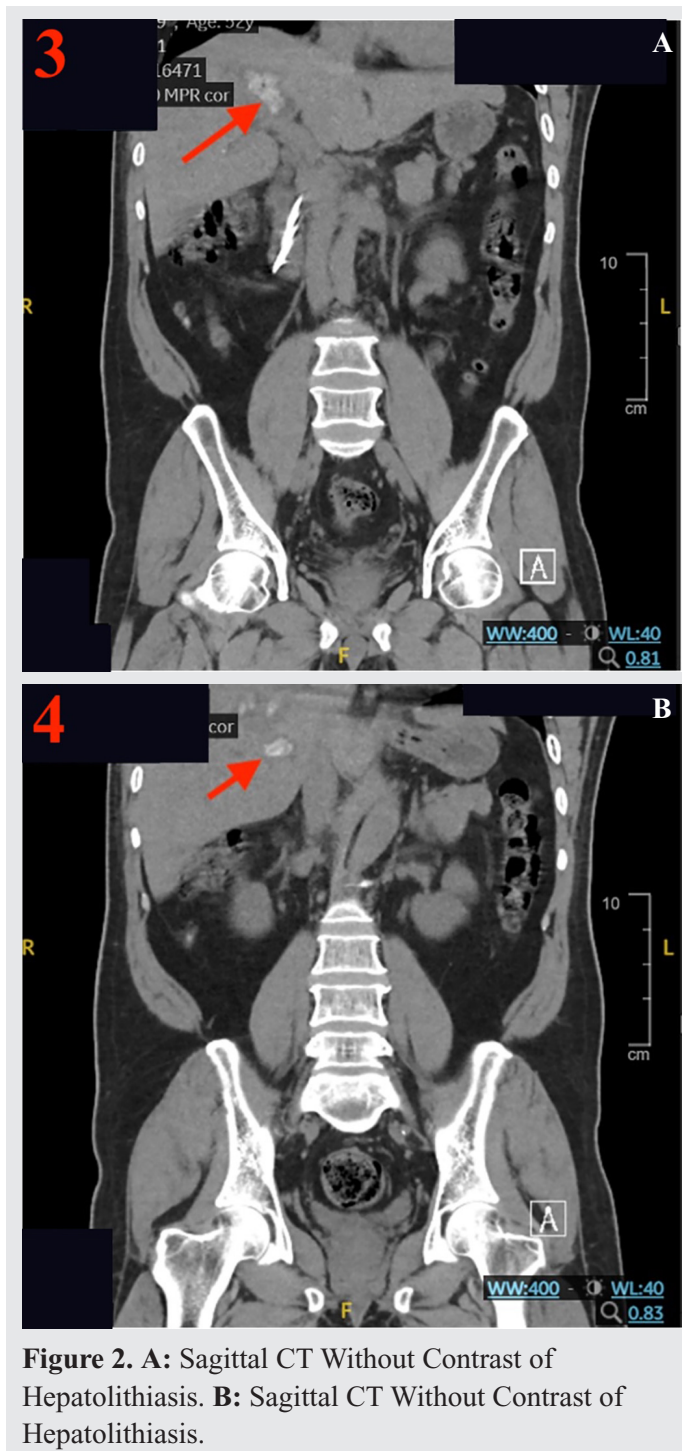
Given his history and imaging findings, the patient was offered a right hepatectomy and biliary drainage procedure. In the operating room, an intraoperative cholangiogram was done, which identified multiple filling defects (indicative of stones). Formal right hepatectomy was performed followed by common bile duct exploration. Stones were evacuated by flushing and suctioning the duct, helping to relieve biliary obstruction. Choledochoscopy was performed to ensure all stones were removed and to confirm no further stones were present. Due to the lithogenic nature of the patient's bile, a procedure to facilitate bile drainage was necessary. Roux-en-y hepaticojejunostomy was performed



**Figure 1. A:** Transverse CT Without Contrast of Hepatolithiasis. **B:** Transverse CT Without Contrast of Hepatolithiasis.

and reconstructed in a side to side fashion with preservation of the common bile duct continuity with the duodenum to allow access to future ERCP if needed.

The patient's postoperative course was straightforward. He was stepped down from the ICU on postoperative day 1, advanced to a regular diet on postoperative day two, and discharged home on postoperative day 3 after all drains were removed. The patient was seen



in the clinic two weeks later and was doing well. A follow-up visit was scheduled three months later alongside a repeat CT, which showed no evidence of recurrence of hepatolithiasis.

## DISCUSSION

Hepatolithiasis is prevalent in East Asia while its incidence in Western countries is considerably lower.<sup>6</sup> The condition is associated with significant morbidity due to recurrent episodes of cholangitis and potential progression to secondary biliary cirrhosis.

The incidence of hepatolithiasis varies widely, with a significantly higher prevalence in regions with endemic liver fluke infections. A review of the literature indicates fewer than 500 cases reported globally, with the majority originating from East Asia. In regions like Taiwan, the reported prevalence is 53.5%, while in Hong Kong it was 3.1% and in Singapore 1.7%, according to a retrospective study of patients between the years 1976–1980.<sup>6</sup>

Patients with hepatolithiasis often present with recurrent episodes of biliary colic, jaundice, and cholangitis. These recurrent episodes can significantly impair the patient's quality of life and pose substantial health risks. Complications associated with hepatolithiasis include liver abscesses, secondary biliary cirrhosis, and, in some cases, cholangiocarcinoma.

Risk factors for hepatolithiasis include conditions that lead to bile stasis, such as biliary strictures or cysts, that significantly increase the risk of stone formation. In addition, recurrent bacterial infections can promote stone formation through chronic inflammation and alterations in bile composition. Parasitic infections, such as clonorchiasis and recurrent pyogenic cholangitis, are particularly implicated in endemic regions. Congenital or acquired anomalies of the biliary tree can predispose individuals to stone formation. These anomalies can create areas of bile stasis and promote chronic inflammation. Dietary habits have been implicated in the risk of hepatolithiasis. Diets high in carbohydrates and low in fats may contribute to bile composition changes that favor stone formation.

Managing refractory hepatolithiasis is challenging due to the high recurrence rates and the complex nature of the condition. The patient in this case had multiple admissions for obstructive jaundice and ascending cholangitis, necessitating repeated ERCP procedures and sphincterotomies. Despite these interventions, intrahepatic stones persisted, highlighting the refractory nature of the condition.<sup>7</sup>

In this case, the decision to perform a formal anatomical hepatectomy and biliary drainage procedure was made based on the extent of intrahepatic lithiasis confined to the right hepatic lobe and the nature of the disease with multiple recurrences.<sup>8</sup>

A review of similar cases in the literature highlights the variability in treatment outcomes for hepatolithiasis. The use of advanced imaging techniques, meticulous surgical planning, and multidisciplinary care are crucial in managing these patients. Future research should focus on identifying genetic markers and novel therapeutic strategies to prevent stone recurrence and improve patient outcomes.

## CONCLUSION

While hepatolithiasis remains a significant clinical challenge, especially in refractory cases, advances in surgical techniques and a multidisciplinary approach can lead to successful management and improved patient outcomes. This case highlights the effectiveness of formal hepatectomy and hepaticojejunostomy in resolving persistent intrahepatic stones when other interventions fail. Continued research and innovation in this field are essential to further enhance the treatment strategies and quality of life for patients suffering from this condition.

**Article citation:** Pendse R, Firouzbakht S, Elfedaly A, Soliman B, Elfedaly M. Surgical management of refractory hepatolithiasis. *The Southwest Journal of Medicine* 2025;13(55):19–22

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**Submitted:** 2/1/2025

**Accepted:** 4/1/2025

**Conflicts of interest:** none

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