



## Case Report

# Diagnosis and management of hepatolithiasis in an adult patient: A case report

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## ABSTRACT

Hepatolithiasis or intrahepatic calculi are common in South East Asia but are rare in Western nations. The primary symptom of the condition is recurrent pain in the upper abdomen. Stones in the cystic duct or common bile duct are also common findings. Recurrent pyogenic cholangitis is the most frequent complication. Radiological studies and percutaneous procedures are vital for diagnosing and managing this condition. The primary goal in treating the condition is to decrease the chance of developing cholangitis and to stop the progression of the disease, which may lead to biliary cirrhosis.

## 1. Introduction and importance

Hepatolithiasis is characterized by the presence of gallstones in all bile ducts peripheral to the confluence of the left and the right hepatic ducts, regardless of the existence of gallstones in other regions of the biliary system, such as the extrahepatic bile duct and the gallbladder [1]. The factors that can lead to hepatolithiasis include the history of recurrent cholangitis, biliary stricture, hepatic abscess, liver atrophy, liver cirrhosis, and a poor prognosis in intrahepatic cholangiocarcinoma [2]. In East Asia, hepatolithiasis is a frequent condition [1,3]. Though symptoms of the intrahepatic kind are more common in younger age groups, the incidence is more in the fifth and sixth decades [4]. Hepatic resection and percutaneous transhepatic cholangioscopic lithotomy (PTCSL) are the primary therapeutic options for hepatolithiasis [5,6]. This case report follows all the SCARE and PROCESS criteria [7,8].

## 2. Case presentation

A 38-year-old female with a history of Autoimmune hepatitis and open cholecystectomy 19 years back for symptomatic gallstone was presented with a recurrent episode of right upper abdominal pain. The pain was insidious on the onset, dull, aching, intermittent, and non-radiating. She didn't give any history of fever, nausea, vomiting, diarrhea or jaundice. She had normal bowel and bladder habits. She had

similar abdominal pain six months back, and endoscopic ultrasound (EUS) showed a common bile duct (CBD) stone (Fig. 1). There is a family history of gallstones. She has been under corticosteroid for 14 years for Autoimmune Hepatitis. She subsequently underwent endoscopic retrograde cholangiopancreatography (ERCP) for CBD stone removal (Fig. 2).

On examination, she was moderately built and oriented to time, place, and person, and her vitals were stable. There were no signs of pallor, icterus, clubbing, cyanosis, or dehydration. On Abdominal examination, there was around 3cm of a surgical scar on the right upper quadrant, soft, non-distended, with mild tenderness present in the right upper quadrant. Blood analysis revealed an increase in alanine aminotransferase (ALT/SGPT) 189U/L, aspartate aminotransferase (AST/SGOT) 219U/L, and alkaline phosphatase (ALP) 407U/L. Magnetic Resonance Cholangiopancreatography (MRCP) which was a multislice two-dimensional (2D) fast-spin-echo showed findings suggestive of hepatolithiasis in the left hepatic duct with dilatation of the left hepatic duct distal to hepatolithiasis (Fig. 3).

She was operated on for a left hepatectomy (left lateral segmentectomy) by a GI surgeon at a tertiary level hospital. The intraoperative assessment showed coarse liver adhesion from previous surgery. Intraoperative contact ultrasonography was done, which did not reveal any calculi in the right hepatic duct and the CBD. The postoperative period was uneventful, and she was discharged on the 4th postoperative day. The postoperative histopathology report showed autoimmune hepatitis

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Fig. 1. A stone of 7.7mm seen in the distal part of CBD. The CBD measures 6.1mm distally and the CHD is mildly dilated at 9.1mm.

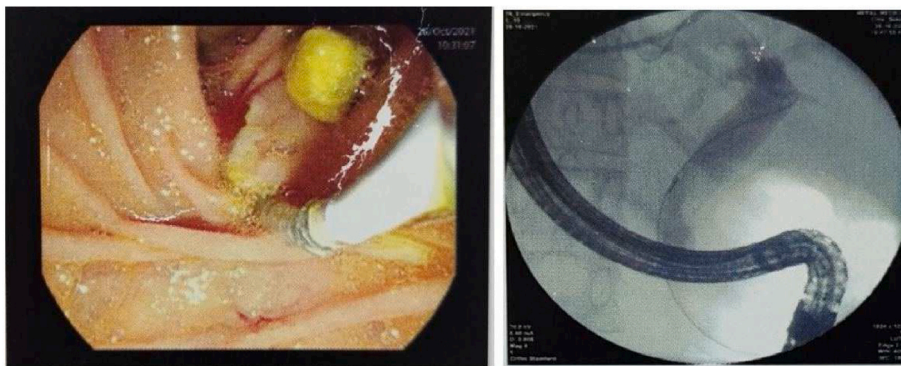


Fig. 2. Cholangiogram showing a 15mm dilated CBD with a 7mm stone in the mid CBD. The intrahepatic ducts are mildly dilated.



Fig. 3. MRCP showing hepatolithiasis in the left hepatic duct with dilatation of the left hepatic duct distal to hepatolithiasis

with liver cirrhosis and sclerosing cholangitis.

### 3. Clinical discussion

There are two forms of intrahepatic stones: primary stones, which are generated in the intrahepatic bile duct, which are more frequent in East Asian countries, and secondary stones developed in the gallbladder, which are common in Western countries [4]. Primary intrahepatic stones have complicated pathogenesis. Intrahepatic stone formation is most commonly associated with bile stasis caused by postoperative strictures, sclerosing cholangitis, Caroli's disease, or neoplasms that result in biliary stenosis and stasis [9,10]. The combination of bile stasis, bile infection, malnutrition, and parasite infestation is likely to cause these stones [11,12]. Proliferative cholangitis (linked to ~ 75% of hepatolithiasis cases in Asia) plays an essential role in the pathogenesis of hepatolithiasis and contributes to high rates of biliary restenosis that cause chronic inflammation necessary to produce intramural and extramural peribiliary gland proliferation [13,14]. Notably, hepatolithiasis is more common in the left lobe

because the left hepatic duct coalesces with the CBD at an acute angle which tends to induce bile stasis when associated with a biliary stricture [4]. Recurrent cholangitis, biliary stricture, hepatic abscess, liver atrophy, liver cirrhosis, and a poor prognosis in intrahepatic cholangiocarcinoma are all related to hepatolithiasis [2].

Asymptomatic patients exist in whom hepatolithiasis is an incidental finding in abdominal imaging [15]. Typical presenting symptoms involve abdominal discomfort, fever, and jaundice, but cholangiocarcinoma is a long-term adverse consequence of the condition. Primary stones are more prevalent in Eastern patients than secondary stones, and recurring pyogenic cholangitis is the most well-known clinical symptom of the disease [16].

Efforts at diagnosing hepatolithiasis should be aimed at accurately locating stones, biliary strictures, and segments of the liver involved and excluding concomitant cholangiocarcinoma [15]. Computed tomography (CT) and abdominal ultrasonography (USG) scans are the principal imaging modalities for hepatolithiasis. Magnetic resonance imaging (MRI) and magnetic resonance cholangiopancreatography (MRCP) can

produce more detailed pictures of the bile duct and detect stones without exposing the patient to radiation [17]. In obstructive jaundice, magnetic resonance cholangiopancreatography (MRCP) has accuracies of 96–100% and 90% for the level and cause of obstruction, respectively, as well as sensitivity, specificity, and accuracy of 97%, 99%, and 98%, respectively, in detecting and locating intrahepatic stones [18].

Hepatolithiasis care is complex, including interdisciplinary treatment to remove stones and bile stasis. The best management for intrahepatic stones is not yet known; however, various surgical and noninvasive methods are available. Nonetheless, the primary aim should be the total removal of stones and eradication of biliary stasis and infection [19]. Non-surgical therapy options for hepatolithiasis include percutaneous transhepatic cholangioscopy lithotripsy, and surgical management includes hepatectomy [14,17,20].

Regardless of treatment modality, these individuals have a high rate of residual stones [5]. The surgical approach to hepatolithiasis involves the removal of the affected segment(s). Hepatectomy removes stones, eliminates strictures and the consequent bile stasis, which causes stone formation, and eradicates the risk of cholangiocarcinoma; in rare cases, it removes a known intraductal tumor [15].

#### 4. Conclusion

Hepatolithiasis is quite common in the 5th and 6th decade, primarily seen in South East Asia; however, in our case, the presentation is in the 3rd decade with a known case of autoimmune hepatitis and sclerosing cholangitis with a history of open cholecystectomy and CBD stone removal. It is essential to practice an interdisciplinary approach to best treat and manage these patients. We start with appropriate imaging and a diligent review of imaging studies with radiologists and surgeons before choosing the optimal treatment path. Early diagnosis is crucial; treatment should be multidisciplinary and complete to avoid recurrence and complications.

#### Ethical approval

Exempted by our institution.

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None declared.

#### Author contributions

Lukash Adhikari: involved in patient management, data collection, design of study, manuscript writing and revision.

Eliz Achhami: design of study, data collection, evidence collection, manuscript writing and revision, corresponding author.

Nabin Bhattarai: design of study, evidence collection, manuscript revision.

Ashim Kandel: design of study, data collection, manuscript revision

Abhigan Babu Shrestha: design of study, manuscript revision.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Guarantor

The Guarantor is the one or more people who accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish Eliz Achhami.

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#### Declaration of competing interest

The authors declare that they have no conflicts of interest.

#### Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2022.104788>.

#### References

- [1] F. Nakayama, Intrahepatic calculi: a special problem in East Asia, *World J. Surg.* 6 (1982) 802–804, <https://doi.org/10.1007/BF01655381>.
- [2] I. Endo, R. Matsuyama, R. Mori, H. Shimada, Intrahepatic Stones, *Blumgart's Surg. Liver*, vols. 1–2, Biliary Tract Pancreas Sixth, 2017, pp. 642–655, <https://doi.org/10.1016/B978-0-323-34062-5.00039-X>, e3.
- [3] F. Nakayama, A. Koga, Hepatolithiasis: present status, *World J. Surg.* 8 (1984) 9–14, <https://doi.org/10.1007/BF01658357>.
- [4] S. Tazuma, Gallstone disease: epidemiology, pathogenesis, and classification of biliary stones (common bile duct and intrahepatic), *Best Pract. Res. Clin. Gastroenterol.* 20 (2006) 1075–1083, <https://doi.org/10.1016/j.BPG.2006.05.009>.
- [5] Y.K. Cheon, Y.D. Cho, J.H. Moon, J.S. Lee, C.S. Shim, Evaluation of long-term results and recurrent factors after operative and nonoperative treatment for hepatolithiasis, *Surgery* 146 (2009) 843–853, <https://doi.org/10.1016/j.SURG.2009.04.009>.
- [6] K. Otani, S. Shimizu, K. Chijiwa, T. Ogawa, T. Morisaki, A. Sugitani, K. Yamaguchi, M. Tanaka, Comparison of treatments for hepatolithiasis: hepatic resection versus cholangioscopic lithotomy, *J. Am. Coll. Surg.* 189 (1999) 177–182, [https://doi.org/10.1016/S1072-7515\(99\)00109-X](https://doi.org/10.1016/S1072-7515(99)00109-X).
- [7] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CASE REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230, <https://doi.org/10.1016/j.IJJSU.2020.10.034>.
- [8] R.A. Agha, C. Sohrabi, G. Mathew, T. Franchi, A. Kerwan, N. O'Neill, A. Thoma, A. J. Beamish, A. Noureldin, A. Rao, B. Vasudevan, B. Challacombe, B. Perakath, B. Kirshtein, B. Ekser, C.S. Pramesh, D.M. Laskin, D. Machado-Aranda, D. Pagano, G. Roy, H. Kadioglu, I.J. Nixon, I. Mukhejee, J.A. McCaul, J. Chi-Yong Ngu, J. Albrecht, J.G. Rivas, K. Raveendran, L. Derbyshire, M.H. Ather, M.A. Thorat, M. Valmasoni, M. Bashashati, M. Chalkoo, N.Z. Teo, N. Raison, O.J. Muensterer, P. J. Bradley, P. Goel, P.S. Pai, R.Y. Affif, R.D. Rosin, R. Coppola, R. Klappenbach, R. Wynn, S. Surani, S. Giordano, S. Massarut, S.G. Raja, S. Basu, S.A. Enam, T. G. Manning, T. Cross, V.K. Karanth, Z. Mei, The PROCESS 2020 guideline: updating consensus preferred reporting of CasE Series in Surgery (PROCESS) guidelines, *Int. J. Surg.* 84 (2020) 231–235, <https://doi.org/10.1016/j.IJJSU.2020.11.005>.
- [9] B. Kayhan, M. Akdoğan, E. Parlak, E. Özarslan, B. Şahin, Hepatolithiasis, *A Turkey experience, Turk. J. Gastroenterol.* 18 (2007) 28–32.
- [10] Y. Ono, K. Kaneko, Y. Ogura, W. Sumida, T. Tainaka, T. Seo, H. Ando, Endoscopic resection of intrahepatic septal stenosis: minimally invasive approach to manage hepatolithiasis after choledochal cyst excision, *Pediatr. Surg. Int.* 22 (2006) 939–941, <https://doi.org/10.1007/S00383-006-1756-Y>.
- [11] H.S. Kaufman, T.H. Magnuson, K.D. Lillemoe, P. Frasca, H.A. Pitt, The role of bacteria in gallbladder and common duct stone formation, *Ann. Surg.* 209 (1989) 584–592, <https://doi.org/10.1097/0000658-198905000-00011>.
- [12] F.M. Cetta, Bile infection documented as initial event in the pathogenesis of brown pigment biliary stones, *Hepatology* 6 (1986) 482–489, <https://doi.org/10.1002/HEP.1840060327>.
- [13] J. Shoda, N. Tanaka, T. Osga, Hepatolithiasis—epidemiology and pathogenesis update, *Front. Biosci.* 8 (2003), <https://doi.org/10.2741/1091>.
- [14] K. Uchiyama, H. Onishi, M. Tani, H. Kinoshita, M. Ueno, H. Yamaue, Indication and procedure for treatment of hepatolithiasis, *Arch. Surg.* 137 (2002) 149–153, <https://doi.org/10.1001/ARCHSURG.137.2.149>.
- [15] S.V. Sakpal, N. Babel, R.S. Chamberlain, Surgical management of hepatolithiasis, *HPB* 11 (2009) 194, <https://doi.org/10.1111/J.1477-2574.2009.00046.X>.

- [16] T. Tsunoda, R. Tsuchiya, N. Harada, R. Yoshino, T. Noda, K. Izawa, T. Yamaguchi, K. Yamamoto, Long-term results of surgical treatment for intrahepatic stones, *Jpn. J. Surg.* 15 (1985) 455–462, <https://doi.org/10.1007/BF02470091>.
- [17] H.J. Kim, J.S. Kim, M.K. Joo, B.J. Lee, J.H. Kim, J.E. Yeon, J.J. Park, K.S. Byun, Y. T. Bak, Hepatolithiasis and intrahepatic cholangiocarcinoma: a review, *World J. Gastroenterol.* 21 (2015), 13418, <https://doi.org/10.3748/WJG.V21.I48.13418>.
- [18] T. Mori, M. Sugiyama, Y. Atomi, Gallstone disease: management of intrahepatic stones, *Best Pract. Res. Clin. Gastroenterol.* 20 (2006) 1117–1137, <https://doi.org/10.1016/J.BPG.2006.05.010>.
- [19] G. Nuzzo, G. Clemente, I. Giovannini, A.M. De Rose, M. Vellone, G. Sarno, D. Marchi, F. Giuliani, Liver resection for primary intrahepatic stones: a single-center experience, *Arch. Surg.* 143 (2008) 570–573, <https://doi.org/10.1001/ARCHSURG.143.6.570>.
- [20] X.D. Wen, T. Wang, Z. Huang, H.J. Zhang, B.Y. Zhang, L.J. Tang, W.H. Liu, Step-by-step strategy in the management of residual hepatolithiasis using post-operative cholangioscopy, *Therap. Adv. Gastroenterol.* 10 (2017) 853–864, <https://doi.org/10.1177/1756283X17731489>.