

e-ISSN 2723-7494

Volume 51 Nomor 1 (2024)

The background of the cover features a blue-tinted photograph of a surgical team in an operating room. A circular inset on the left shows a close-up of a surgical incision. Large, semi-transparent yellow and blue circles are overlaid on the image. The title is centered in a large, bold, sans-serif font.

JURNAL ILMU BEDAH INDONESIA

Diterbitkan oleh:

Ikatan Ahli Bedah Indonesia

HEPATOLITHIASIS WITH CONCOMITANT SPINE SCHWANNOMA IN 44-YEAR-OLD INDONESIAN WOMAN IN REFERRAL HOSPITAL, NORTH SUMATRA, INDONESIA

¹Safruddin Nasution, ²Habib Al Kahfi Nasution, ²Sayyid Muhammad Ziyaad

¹Division of Digestive Surgery, Department of Surgery, Faculty of Medicine, Universitas Sumatera Utara, Medan, Indonesia

²Medical Doctor, Faculty of Medicine, Universitas Sumatera Utara, Medan, Indonesia

Introduction: Hepatolithiasis, a benign bile disease prevalent in Asia, and spinal schwannoma, a rare nerve sheath tumor, have never been reported concomitantly.

Case Presentation: This report presents a rare case of a 44-year-old Indonesian woman with hepatolithiasis and concomitant spinal schwannoma, emphasizing the importance of collaborative treatment strategies. The patient exhibited abdominal pain, jaundice, and neurological symptoms, prompting surgical interventions including left hepatectomy for hepatolithiasis and tumor excision for spinal schwannoma.

Discussion: Prompt treatment and post-operative management leads to significant improvement in symptoms.

Conclusion: This case underscores the necessity of early recognition, invasive treatment, and rehabilitative therapy to mitigate complications and enhance patient outcomes.

Keywords: Hepatolithiasis; Spinal Schwannoma; Surgery management

INTRODUCTION

Hepatolithiasis is a type of benign bile disease, characterized by either complete or partial obstruction in biliary hepatic duct, is considered to be an endemic disease in Asian.¹ In Europe and American, the incidence of it increases. Otherwise, The global Incidence of Hepatolithiasis has also increased from 0.32/100.000 To 0.85/100.000 within the decades.²⁻⁴

Spinal scwannoma is regarded as a rare disease, With an incidence of 0.3-0.5/100.000 annually.⁵ Schwannoma is considered the most common of nerve sheat tumor.^{6,7}

Simultaneous occurrence of Hepatolithiasis and Spinal schwannoma has never been reported. This Case Report aim to report rare case and elicit considerable decision making for treating the patient with Hepatolithiasis with concominant spine schwannoma, which happened in North Sumatra, Indonesia.

CASE PRESENTATION

A 44-year-old Indonesian Bataknese farmer woman was referred from primary health care with intermittent pain on the right-upper quadrant of abdomen for the last 6 month to the surgery clinic in Adam Malik referral hospital. The pain was relatively mild and described as localized but blunt, in which the patient reported a significant intensity increase for the last month. The patient experienced pain the most at the night time.

The patient reported brownish urine, similar to a concentrated tea. She also experienced a difficulty in defecating within the same onset although the symptoms had been reduced significantly since a week treatment of stool laxative. She had consumed anthelmintic agents for a month by the primary healthcare without evidence of stool analysis. She doesn't routinely use either footwear or gloves. On physical examination, she appeared generally stable, apart from several remarkable findings such as bilateral icteric sclera, hepatomegaly, positive Murphy's sign.

During the two previous months, the patient also noticed a progressive weakness bilaterally and symmetrically on the lower extremities; as accompanied by symmetrical numbness starting from mid-abdominal level, and mostly perceived on the both lower extremities. She had experienced radiating pain on her waist to both lower limbs. The patient was refered to the orthopaedic surgeon to be examined. Evaluation of the patient's both lower extremities revealed significant motoric weakness and hypoesthesia. Moreover, we also found a positive pathological reflexes on the right side.

To assess the abdominal symptoms, we explore the patients' anatomical condition by whole abdominal multi-slice CT-scan with IV oral and anal contrast. We found a dilated both of intrahepatic and extrahepatic biliary duct, multiple hyperdense mass in intrahepatic biliary duct. The expert concluded a finding of hepatomegaly and hepatolithiasis (with Dong's classification type II A) (figure 1). We

performed an open left hepatectomy procedure to dissect whole left lobe of the liver. Post-operative findings were presented in Figure 2, as we found multiple solid and blackish-appeared stones of various sizes ($\pm 0.5 - 2$ cm in diameter). The patient's abdominal symptoms quickly receded after hepatectomy surgery.

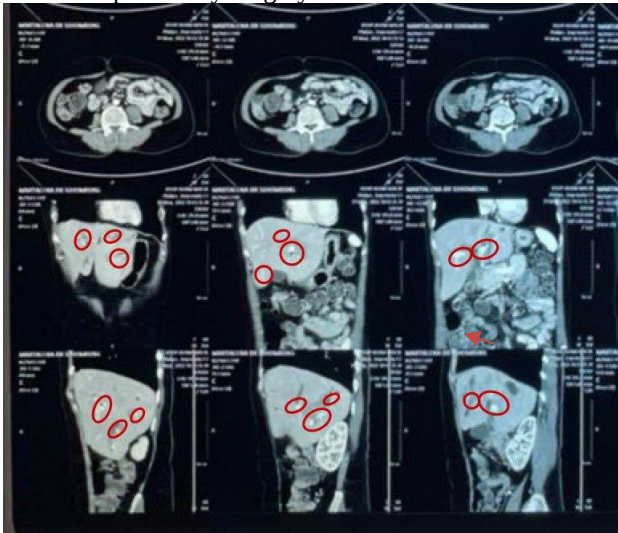


Figure 1. MSCT imaging with oral and anal contrast revealed a significant amount of hepatic stones on the left liver lobe (on the red markers).



Figure 2. Post-operative findings of the patient's resected liver lobe and 10 found stones in total with the diameter of 1 to 2 cm per stone.

To assess neurological symptoms, A whole spine MRI assessment without contrast showed space-occupying-lesion (SOL) intradural-extramedullary within T7-T8 (Frenkel type B), elliptically-shaped and $\pm 1.5 \times 3.0$ cm in size, compressing the right medulla toward the left side, markedly defined, and no additional SOL was suspected on the other medullary level (figure 3). Histopathological analysis revealed a Schwannoma finding of the investigated tissue, with no apparent finding of malignant transformation. Tumor excision surgery was performed one month after hepatectomy, and observe a significant improvement of motoric strength to almost normally functioned lower extremities after a month. After 2 months of undergoing series of rehabilitative therapy, The patient symptoms has been fully receded.

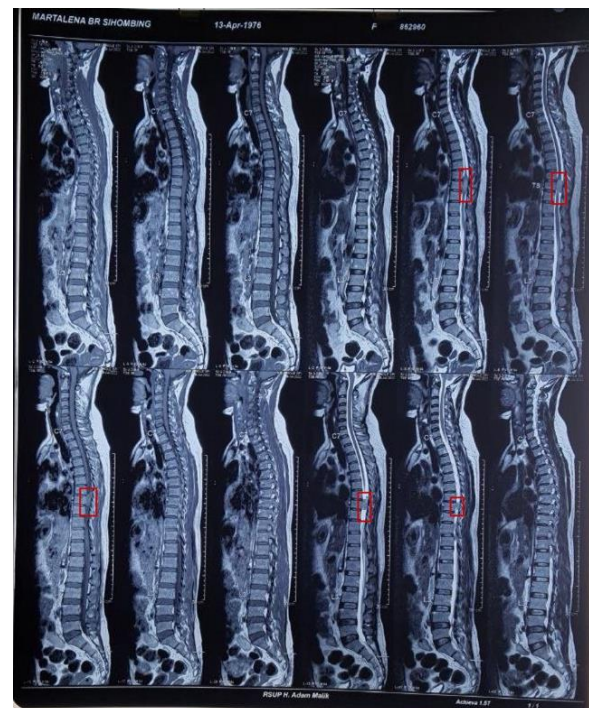


Figure 3. Pre-operative imaging by MRI of the whole spine on sagittal show SOL within the T7-T8 (on the red markers).



Figure 4. Pre-operative imaging by MRI of the whole spine on Axial show SOL within the T7-T8 (On the red markers).

DISCUSSIONS

This case report illustrates a rare case of hepatolithiasis concomitant with spine schannoma. Even though both of them are separately different entities, this concurrence presentation has never been reported yet. This patient presents common bile duct coexisting with intrahepatic bile-duct stone.^{8,9} This patient presents intrahepatic and extrahepatic bile stone, the stone is located in intrahepatic biliary duct, without stenosis of bile duct, with slight dilatation, which is rare.^{10,11}

This case report presented high hepatolithiasis risk factor including low socioeconomic status, living in the rural countryside which the well water isn't available properly, having history of parasitic

infestation, and barefoot walker.¹²

Invasive treatment is necessary to prevent further complications such as cholangitis suppurative, septicemia, secondary cirrhosis hepatic, portal hypertension, varices bleeding, and hepatic failure.^{13,14} The left hepatectomy was chosen because it has lower complication and effective for either bilateral hepatolithiasis (Type I) or left hepatolithiasis (Type L).¹⁵ The availability facilities and the dilatation of the ductus hepatic because of the stone presentation are considered to perform left hepatectomy.

Spinal schwannoma are rare disease, prevailing studies accounted only for 2% of spinal tumor.¹⁶ The schwannoma tumor generally happened asymptomatic, these tumors generally identified unintentionally during studies for other medical reason.^{16,17} Whereas, In our patient presented with progressive lower extremities weakness and sensory loss, which are rare. While occurrence intradural extramedullary within thoracic region in our patient is not common. Spinal schwannoma tumors mostly happen in lumbar region.¹⁸

Tumor excision was performed one month after hepatectomy. The timing gap was considered because of the patient stabilization after left hepatectomy. Surgical resection using posterior approach through laminectomy was chosen because it is the gold standard for treatment of spinal schwannoma.¹⁹ Rehabilitative therapy have been conducted for the patient to be independent with post op precaution.^{20,21}

CONCLUSION

To conclude our report, this case presented first concurrence of hepatolithiasis and spinal schwannoma even though both of them are separated entities. Early recognition, invasive treatment, and rehabilitative therapy of these diseases can prevent further complication and improve patient's quality of life.

REFERENCES

1. Feng X, Zheng S, Xia F, et al. Classification and management of hepatolithiasis: A high-volume, single-center's experience. *Intractable rare Dis Res* 2012; 1: 151–156.
2. Mori T, Sugiyama M, Atomi Y. Gallstone disease: Management of intrahepatic stones. *Best Pract Res Clin Gastroenterol* 2006; 20: 1117–1137.
3. Lubezky N, Facciuto M, Harimoto N, et al. Surgical treatment of intrahepatic cholangiocarcinoma in the USA. *J Hepatobiliary Pancreat Sci* 2015; 22: 124–130.
4. Kim HJ, Kim JS, Joo MK, et al. Hepatolithiasis and intrahepatic cholangiocarcinoma: A review. *World J Gastroenterol* 2015; 21: 13418–13431.
5. Seppälä MT, Haltia MJ, Sankila RJ, et al. Long-term outcome after removal of spinal schwannoma: a clinicopathological study of 187 cases. *J Neurosurg* 1995; 83: 621–626.
6. Jinnai T, Hoshimaru M, Koyama T. Clinical Characteristics of Spinal Nerve Sheath Tumors: Analysis of 149 Cases. *Neurosurgery*; 56.
7. Safavi-Abbasi S, Senoglu M, Theodore N, et al. Microsurgical management of spinal schwannomas: evaluation of 128 cases. *J Neurosurg Spine* 2008; 9: 40–47.
8. Jindal S, Bagwan AI, Rathinasamy R, et al. Hepatolithiasis: A Retrospective Analysis of Surgical Management Options in a Tertiary Care Centre in Southern India. *Cureus* 2022; 14: e27797.
9. Tazuma S. Gallstone disease: Epidemiology, pathogenesis, and classification of biliary stones (common bile duct and intrahepatic). *Best Pract Res Clin Gastroenterol* 2006; 20: 1075–1083.
10. Nakayama F. Intrahepatic calculi: a special problem in East Asia. *World J Surg* 1982; 6: 802–804.
11. Uchiyama K, Onishi H, Tani M, et al. Indication and Procedure for Treatment of Hepatolithiasis. *Arch Surg* 2002; 137: 149–153.
12. Shoda J, Tanaka N, Osuga T. Hepatolithiasis--epidemiology and pathogenesis update. *Front Biosci* 2003; 8: e398-409.
13. Shah OJ, Robbani I, Shah P, et al. Left-sided hepatic resection for hepatolithiasis: a longitudinal study of 110 patients. *HPB Off J Int Hepato Pancreato Biliary Assoc* 2012; 14: 764–771.
14. Jeng KS, Yang FS, Ohta I, et al. Dilatation of intrahepatic biliary strictures in patients with hepatolithiasis. *World J Surg* 1990; 14: 583–587.
15. Hwang S, Lee S-G, Kim M-H, et al. Intraoperative biliary exploration through the left hepatic duct orifice during left hepatectomy in patients with left-sided hepatolithiasis. *Langenbeck's Arch Surg* 2008; 393: 383–389.
16. Lim D-J. Atypical intradural extramedullary spinal schwannoma causing cauda equina syndrome: A case report and literature review. *International journal of surgery case reports* 2023; 108: 108396.
17. Nadeem M, Mansoor S, Assad S, et al. Spinal Schwannoma with Intradural Intramedullary Hemorrhage. *Cureus* 2017; 9: e1082.
18. Jeon JH, Hwang HS, Jeong JH, et al. Spinal schwannoma; analysis of 40 cases. *J Korean Neurosurg Soc* 2008; 43: 135–138.
19. Lenzi J, Anichini G, Landi A, et al. Spinal Nerves Schwannomas: Experience on 367 Cases - Historic Overview on How Clinical, Radiological, and Surgical Practices Have Changed over a Course of 60 Years. *Neurol Res Int*; 2017. Epub ahead of print 2017. DOI: 10.1155/2017/3568359.
20. Mobbs RJ, Li J, Sivabalan P, et al. Outcomes after decompressive laminectomy for lumbar spinal stenosis: comparison between minimally invasive unilateral laminectomy for bilateral decompression and open laminectomy: clinical article. *J Neurosurg Spine* 2014; 21: 179–186.



21. Low M, Burgess LC, Wainwright TW. A Critical Analysis of the Exercise Prescription and Return to Activity Advice That Is Provided in Patient Information Leaflets Following Lumbar Spine Surgery. *Medicina (Kaunas)*; 55. Epub ahead of print July 2019. DOI: 10.3390/medicina55070347.



This work is licensed under
a Creative Commons Attribution