Radiology Case Reports 20 (2025) 1963–1966

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Ciliated- hepatic cyst: A case report with literature review ☆

*Asmae Guennouni**, *Zaynab Iraqui Houssaini*, *Soukaina Bahha*, *Hassane En-nouali*, *Jamal El Fenni*

Department of Radiology at the Military Hospital, Rabat

ARTICLE INFO

Article history:
Received 21 December 2024
Accepted 1 January 2025

Keywords:
Pseudostratified ciliated epithelium
Imaging
MRI
Ciliated hepatic cyst

ABSTRACT

Ciliated hepatic cyst (CHRC) is a rare, benign hepatic lesion characterized by a pseudostratified ciliated epithelium, distinct from intrahepatic bile ducts. First described in 1984, fewer than 80 cases have been reported. These cysts are often asymptomatic and discovered incidentally during imaging. Radiologically, they can mimic other hepatic lesions, making diagnosis challenging. In this case, a 60-year-old male with right iliac fossa pain was found to have a cyst in segment VIb. MRI suggested a benign lesion, and histopathological analysis after surgical resection confirmed CHRC. Given its potential for malignant transformation, surgical resection is recommended even in the absence of clear signs of progression.

Introduction

Hepatic cysts are common, typically benign lesions that are often asymptomatic and discovered incidentally during imaging studies performed for other conditions. Among these, the ciliated-lining hepatic cyst is an extremely rare entity characterized by the presence of a pseudostratified ciliated epithelium, similar to that of the respiratory tract but distinct from intrahepatic bile ducts. This entity was first introduced in 1984 by Wheeler and Edmondson1 [1], and since then, fewer than 80 cases have been reported in the literature [2].

Radiologically, the ciliated-lining hepatic cyst can be mistaken for other benign or malignant hepatic lesions, making preoperative diagnosis challenging. Imaging plays a crucial role in detecting these cysts, although the definitive diagnosis relies on histopathological examination.

This report highlights the importance of considering this rare pathology in the differential diagnosis of hepatic cystic lesions, especially when localized in the right hepatic lobe, and underscores the need for surgical management due to the potential risk of malignant transformation.

Case report

A 60-year-old male patient with no history of prior surgeries presented with a 2-month history of persistent right iliac fossa

☆ Competing Interests: The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

* Corresponding author.
E-mail address: asmaguennouni95@gmail.com (A. Guennouni).

https://doi.org/10.1016/j.radcr.2025.01.001

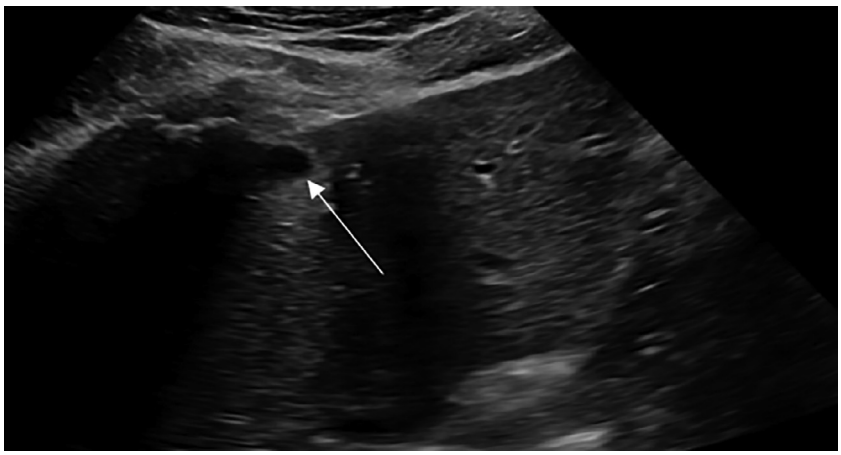

Fig. 1 – A subcapsular cystic lesion in segment IVa with a small hyperechoic sediment (white arrow).

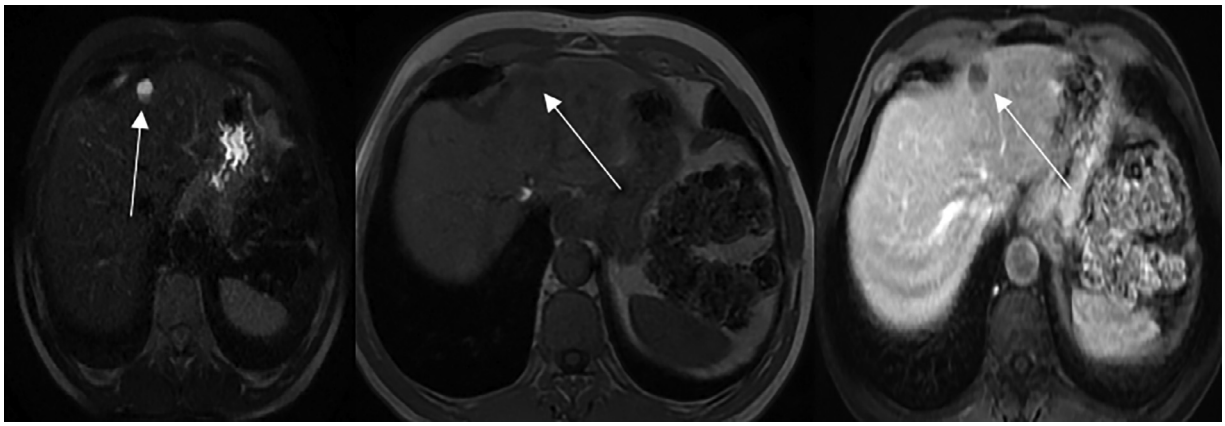

Fig. 2 – A cystic lesion in segment IVa with hyperintense T2 signal and sediment (A), hypointense T1 signal with slightly hyperintense T1 sediment, and no enhancement after gadolinium injection3.

pain. He had a medical history of diabetes mellitus and hypertension, both under treatment. The pain was not accompanied by any other symptoms such as fever, weight loss, or digestive disturbances.

On physical examination, no abnormalities were detected, and the patient appeared clinically stable. Laboratory tests, including a complete blood count, inflammatory markers, and liver function tests, were within normal limits, showing no signs of infection or other systemic involvement.

Initial imaging with abdominal ultrasound revealed a cystic mass located in hepatic segment VIb. The lesion measured approximately 4 × 4 mm and was characterized by a small dependent sediment. No evidence of gallbladder lithiasis was observed. To further evaluate this finding, magnetic resonance imaging (MRI) was performed (Fig, 1).

A magnetic resonance imaging (MRI) examination was performed to better characterize this mass. It appeared hypointense compared to the adjacent hepatic parenchyma on T1-weighted sequences and distinctly and uniformly hyperintense on T2-weighted sequences, with no enhancement after gadolinium injection and no enhancement of the cyst wall.

This mass contained sediment that was slightly hyperintense on T1-weighted sequences and hypointense on T2-weighted sequences. There was no direct communication with the biliary ducts (Fig. 2).

The differential diagnoses were primarily biliary cystadenoma, lymphangioma, and hemorrhagic biliary cyst.

Consequently, in the absence of a definitive etiological diagnosis, an en-bloc resection of the mass was performed. A well-encapsulated mass was identified in segment IVa.

The patient was able to return home three days after the procedure. The postoperative course was uneventful.

Histological examination of the surgical specimen revealed a fibrous wall lined with pseudostratified ciliated columnar epithelium. The diagnosis of a ciliated hepatic cyst was finally established.

Discussion

The ciliated hepatic cyst (CHRC) is a rare, benign, solitary intrahepatic cystic lesion characterized by the presence of

pseudostratified ciliated epithelium, similar to that of the respiratory tract but distinct from intrahepatic biliary epithelium [1,3].

It develops from the foregut within the liver. It results from an abnormal tracheobronchial budding that, after detaching, may migrate caudally to become embedded in the liver before the closure of the pleuroperitoneal canals, which occurs around the end of the eighth week of development [4].

The majority of these cysts are subcapsular and located in the left hepatic lobe.

First described by Wheeler and Edmondson in 1984, CHRC remains an exceptionally rare pathology, with fewer than 80 cases documented in the international literature [2]. Its rarity and often subtle clinical presentation complicate its diagnosis, making it a condition of interest for clinicians and pathologists.

It can be difficult to distinguish CHRC from other hepatic lesions, with differential diagnoses primarily including biliary cystadenoma, lymphangioma, and hemorrhagic biliary cyst.

Clinically, CHRCs are generally asymptomatic and are discovered incidentally during imaging examinations [5]. However, in symptomatic cases, patients may present with localized abdominal pain in the right hypochondrium, or compressive symptoms if the cyst reaches a significant size. These cysts are most often located in the right hepatic lobe, particularly in segment IVa [6]. More rarely, CHRCs may occur in other hepatic segments, including rare locations near the gallbladder in segment IVb, reported in 3.6 % of cases [7].

The radiological diagnosis of CHRC relies on increasingly sophisticated imaging modalities. On ultrasound, CHRCs appear as well-defined, subcapsular, hypoechoic, or anechoic lesions, often accompanied by posterior enhancement. Some cases may show dependent hyperechoic sediment [8]. While these findings may suggest a simple cyst, unusual location or the presence of sediment warrants further investigation.

On computed tomography (CT), CHRCs appear as hypodense, well-defined lesions without enhancement after contrast administration. In some cases, dependent hyperdense sediment may be visualized. MRI is particularly useful in characterizing these cysts. On T2-weighted sequences, CHRCs are strongly hyperintense, while on T1-weighted sequences, their intensity varies, sometimes appearing slightly hyperintense due to intracystic sediment. Unlike more complex lesions such as biliary cystadenomas, CHRCs do not show enhancement after gadolinium administration [9,10]. However, in the absence of mural calcifications, which are rare, it can be challenging to distinguish CHRCs from other hepatic cystic pathologies.

CHRCs can be mistaken for several other hepatic cystic lesions. Differential diagnoses include simple cysts, hydatid cysts, amebic or pyogenic abscesses, intrahepatic biliary cysts, and cystic tumors such as biliary cystadenomas. An integrated approach combining clinical history, biological findings, such as elevated infection markers indicative of a hepatic abscess, and imaging characteristics is essential to narrow down the differential diagnosis. Nevertheless, these elements are often insufficient, requiring histopathological examination for a definitive diagnosis.

CHRCs exhibit a characteristic wall structure composed of 4 layers: an inner layer of pseudostratified ciliated epithelium, a layer of underlying connective tissue, a thin layer of smooth muscle fibers, and an external fibrous capsule [3]. These features distinguish CHRCs from other hepatic cystic lesions. Cytology, obtained via fine-needle aspiration (FNA), may reveal ciliated cells, which are pathognomonic for this lesion [10]. However, due to the low cellularity of the samples, surgical resection is indicated.

The evolution of CHRC is poorly understood. Two cases of compression of neighboring structures have been described: 1 case of compression of the portal vein trunk leading to portal hypertension in an adolescent, with regression of symptoms after cyst resection [11], and 1 case of biliary compression presenting as jaundice [12]. Moreover, 3 cases of malignant transformation into squamous cell carcinoma have been reported. This low risk of malignant transformation and progression to carcinoma underscores the need for a proactive approach, including surgical resection, even in the absence of obvious signs of progression. Furthermore, the benign nature of CHRCs may be misinterpreted as malignancy during preoperative imaging, emphasizing the importance of resection to exclude more serious diagnoses and reduce the risk of future complications.

Conclusion

CLHC, though rare, is a critical diagnostic consideration in the evaluation of hepatic cystic lesions. Its clinical and radiological features aid in distinguishing it from other cystic pathologies, but definitive diagnosis relies on histopathological examination. Given the potential for malignant transformation and the difficulty of excluding other serious conditions, surgical resection remains the treatment of choice. Increased recognition of this entity through advancements in diagnostics contributes to a better understanding of its clinical behavior and optimal management strategies.

Patient consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

REFERENCES

[1] Wheeler DA, Edmondson HA. Ciliated hepatic cyst. Am J Surg Pathol 1984;8(5):467–70.
[2] Horii T, Ohta M, Mori T, Uchida H, Iwata H, Miwa S, et al. Ciliated hepatic cyst: case report and literature review. Hepatol Res 2003;26(4):243–8.
[3] Terada T, Nakanuma Y, Kono N, Saito K, Ikeda S, Ohno A, et al. Ciliated hepatic cyst: hhistochemical and ultrastructural study of three cases compared with normal bronchi and intrahepatic bile ducts. Am J Surg Pathol 1990;14(4):356–63.
[4] Chatelain D, Chailley-Heu B, Terris B, Molas G, Vissuzaine C, Valla D, et al. The ciliated hepatic foregut cyst, an unusual

bronchiolar foregut malformation: a histological, histochemical, and immunohistochemical study of 7 cases. Hum Pathol 2000;31(2):241–6.
[5] Jakowski JD, Lucas JG, Seth S, Burke A, Sobin LH, Badizadegan K, et al. Ciliated hepatic cyst: a rare but increasingly reported hepatic cyst. Ann Diagn Pathol 2004;8(6):342–6.
[6] Ambe C, Gonzalez-Cuyar L, Buchino JJ, McGowan SE, Hsu Y, et al. Ciliated hepatic cyst: a review of 103 cases in the global literature. Open J Pathol 2012;2(1):45–9.
[7] Benlolo D, Vilgrain V, Terris B, Zins M, Paradis V, Belghiti J, et al. Imaging of ciliated hepatic or biliary cysts. Gastroenterol Clin Biol 1996;20(5):497–501.
[8] Kaplan KJ, Escobar M, Vance K, Demicco EG, Keating J, et al. Ciliated hepatic cyst: rreport of a case via fine-needle aspiration. Diagn Cytopathol 2007;35(4):245–9.
[9] Boumoud M, Daghfous A, Maghrebi H, Beji S, Ouerhani R, Djemel F, et al. Imaging aspects of ciliated hepatic cyst. J Radiol Diagn Interv 2015;96:S78–81.
[10] Vick DJ, Goodman ZD, Ishak KG, Baker SP, Johnson EF, Palmer JE, et al. Carcinoma arising in a ciliated hepatic cyst. Arch Pathol Lab Med 1999;123(11):1115–17.
[11] Harty MP, Hebra A, Ruchelli ED, Schnaufer L, Ginsberg JP, Reed ML, et al. Ciliated hepatic foregut cyst causing portal hypertension in an adolescent. AJR Am J Roentgenol 1998;170(3):688–90.
[12] Wheeler DA, Edmondson HA. Ciliated hepatic foregut cyst. Am J Surg Pathol 1984;8(5):467–70.