Ciliated Hepatic Foregut Cyst: A Case-Report and Literature Review

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Abstract

The ciliated hepatic foregut cyst (CHFC) is a rare benign cystic lesion of the liver. Since its initial description fewer than one hundred cases have been reported in the literature. CHFCs are usually asymptomatic and often discovered incidentally during imaging for other reasons. They are most often located in the anterior subcapsular region of segment IV of the liver. The definitive diagnosis is histopathological.

Case Report

We report the case of a 60-year-old man with no significant medical or surgical history who underwent a thoraco-abdomino-pelvic CT scan as part of the workup for a pathological fracture of the left femur. It revealed an oval, well-circumscribed cystic lesion in the anterior subcapsular region of segment IVa. US and MRI were used for further assessment.

Discussion

CHFCs are rare cystic liver lesions resulting from abnormal embryonic development. Although benign, rare cases of malignant transformation have been reported . They typically occur in adults between 50 and 55 years, and a slight male predominance. Most cases are asymptomatic. They are characteristically located in the anterior subcapsular region of segment IV —a key diagnostic clue. Ultrasound, is usually the first-line modality. It reveals a well-defined unilocular cyst with dependent echogenic sediment. CT typically shows a spontaneously hypodense lesion without post-contrast enhancement. MRI is the gold standard for lesion characterization. It reveals the pathognomonic "smurf's head" appearance, as seen in our case. Histological confirmation is recommended. Management remains controversial as CHFC is not purely benign due to its malignant transformation potential.

Conclusion

CHFC is a rare benign liver lesion that should be considered in any middle-aged adult presenting with a unilocular cystic lesion in segment IV.

Keywords: *Ciliated Hepatic Foregut Cyst (CHFC), MRI, Benign Hepatic Cystic Lesion, CT, US.*

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Introduction

The ciliated hepatic foregut cyst (CHFC) is a rare benign cystic lesion of the liver (1, 2). Since its initial description by Friederich in 1857 and its modern designation by Wheeler and Edmondson in 1984 (3), fewer than one hundred cases have been reported in the literature (3, 4). CHFCs are usually asymptomatic (1, 3, 5) and often discovered incidentally during imaging for other reasons. They are most often located in the anterior subcapsular region of segment IV of the liver (1, 6). The definitive diagnosis is histopathological, demonstrating a typical four-layer architecture: ciliated epithelium, connective tissue, smooth muscle, and fibrous capsule (3, 7).

Case Report

We report the case of a 60-year-old man with no significant medical or surgical history who underwent a thoraco-abdomino-pelvic CT scan as part

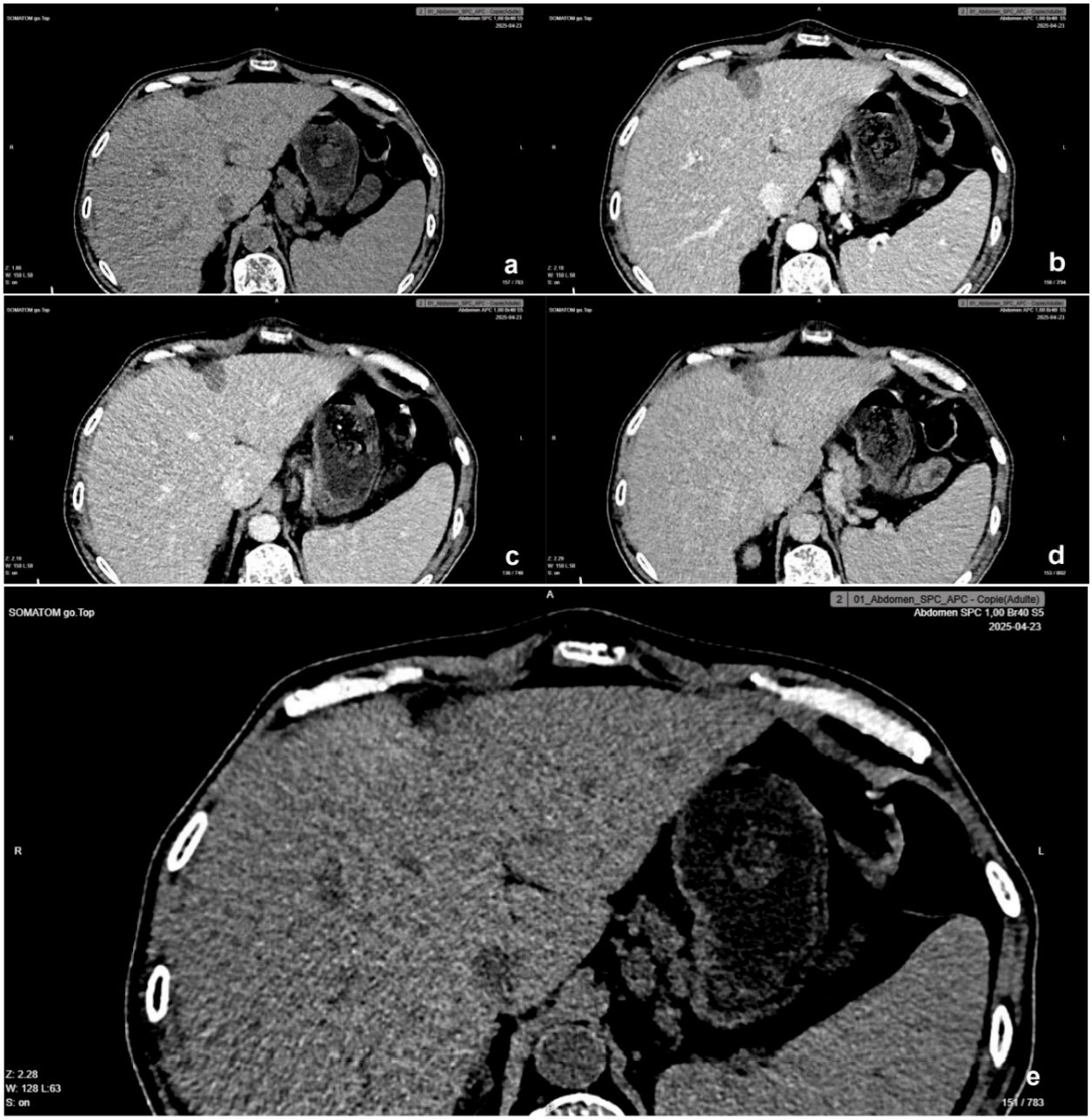

Figure 1. *Axial CT slices without (a, e) and with contrast injection at arterial (b), portal (c), and delayed hepatic (d) phases, showing a rounded, well-circumscribed anterior subcapsular cystic lesion in segment IVa spontaneously hypodense (a) and heterogeneous due to dependent material isodense to the parenchyma (e), without enhancement at any post-contrast phase.*

of the workup for a pathological fracture of the proximal left femur following a fall from standing height.

Multiphasic CT revealed an oval, well-circumscribed cystic lesion in the anterior subcapsular region of segment IVa, with a predominant anteroposterior axis. It appeared spontaneously hypodense and heterogeneous due to a dependent component isodense to the liver parenchyma, without enhancement on any of the post-contrast phases (Fig. 1).

Ultrasound using both low-frequency (3–5 MHz) and high-frequency (6–13 MHz) probes identified a unilocular cystic lesion (anechoic with posterior acoustic enhancement) with the same morphological characteristics as on CT, containing heterogeneous dependent material iso-echoic to the parenchyma, without Doppler signal (Fig. 2).

MRI including T2 HASTE, T2 HASTE FatSat, T2 BLADE respiratory triggered, T1 VIBE DIXON IPOP, T1 VIBE DIXON C+ dynamic, DWI/ADC (b =

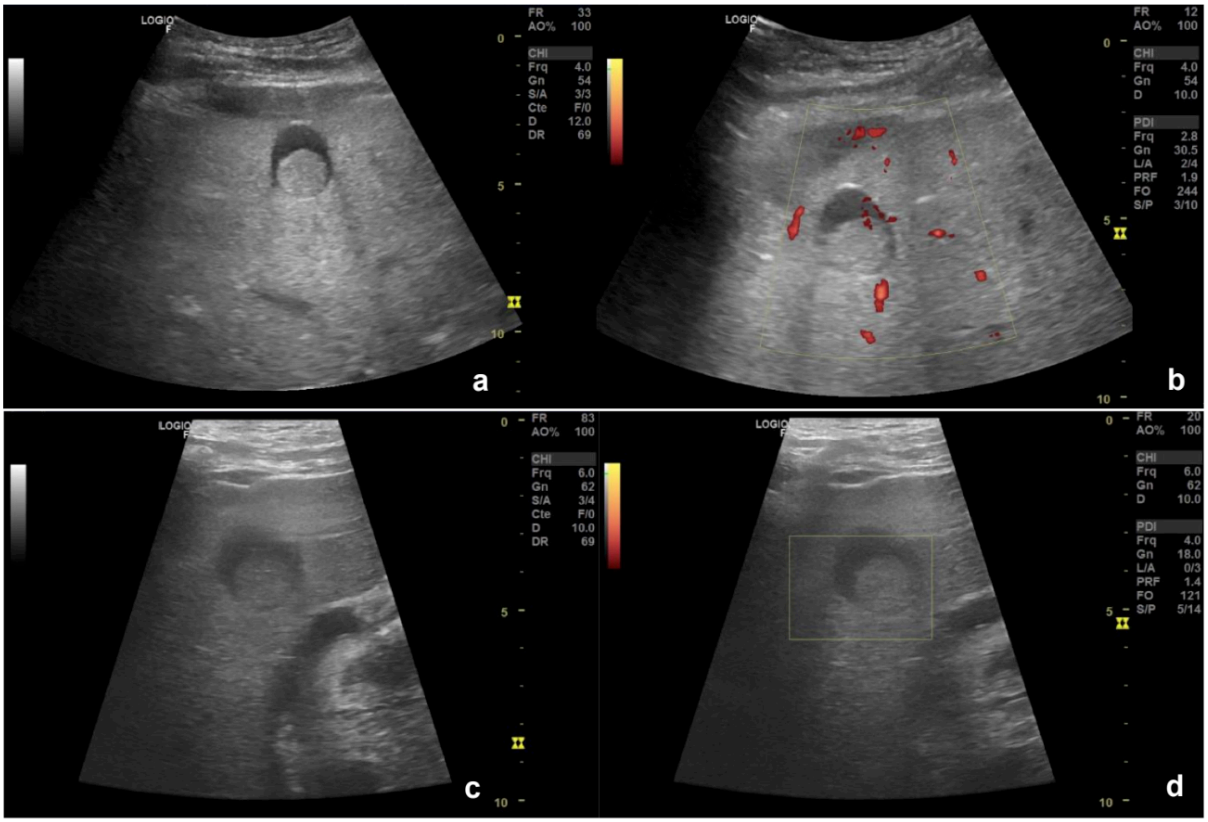

Figure 2. *Transverse ultrasound images of the epigastric region, at low (a, b) and high frequency (c, d) in B mode (a, c) and power Doppler (b, d), showing a well-defined, unilocular aval cystic lesion, anechoic with posterior acoustic enhancement, containing heterogeneous dependent material iso-echoic to the parenchyma, without Doppler signal.*

50, 400, 800) and 3D MRCP sequence was performed for further etiological evaluation. The lesion appeared heterogeneously hyperintense on T2-weighted images and heterogeneously hypointense on T1-weighted images due to the presence of dependent material with intermediate T2 signal and hyperintense T1 signal, producing the characteristic “smurf’s head” appearance on coronal sequences. Diffusion-weighted imaging with multiple b-values showed no diffusion restriction. Post-contrast sequences revealed no enhancement of the cyst contents but demonstrated moderate, delayed mural enhancement (Fig. 3).
Given these typical imaging features, the diagnosis of ciliated hepatic foregut cyst was made.

Discussion

CHFCs are rare cystic liver lesions resulting from abnormal migration of the primitive foregut during embryonic development (1, 2). Although benign, rare cases of malignant transformation into squamous cell carcinoma have been reported (1-4), with an estimated risk of about 3% (8).
They typically occur in adults, with a median age between 50 and 55 years (2, 6), and a slight male predominance (8, 9). Most cases are asymptomatic, as in our patient, but they may occasionally present with right upper quadrant pain, obstructive jaundice, or portal hypertension depending on location.
CHFCs are most often smaller than 30 mm (6) and are characteristically located in the anterior subcapsular region of segment IV (6)—a key diagnostic clue (6, 8).

Ultrasound, usually the first-line modality (4, 6), reveals a well-defined unilocular cyst (6), hypo- or anechoic with posterior enhancement (6, 10). Dependent echogenic sediment is often seen (4, 6). In some cases, the lesion may mimic a solid tumor due to its thick mucinous content (6).

CT typically shows a spontaneously hypodense lesion without post-contrast enhancement (6, 10), frequently with hyperdense sediment (4, 6). Rarely, the lesion may be spontaneously hyperdense (6, 10).

MRI is the gold standard for lesion characterization (6, 11, 12). In more than 50% of cases, the cyst appears hyperintense on T1-weighted images due to protein-rich mucinous content (6, 11), and intensely hyperintense on T2-weighted

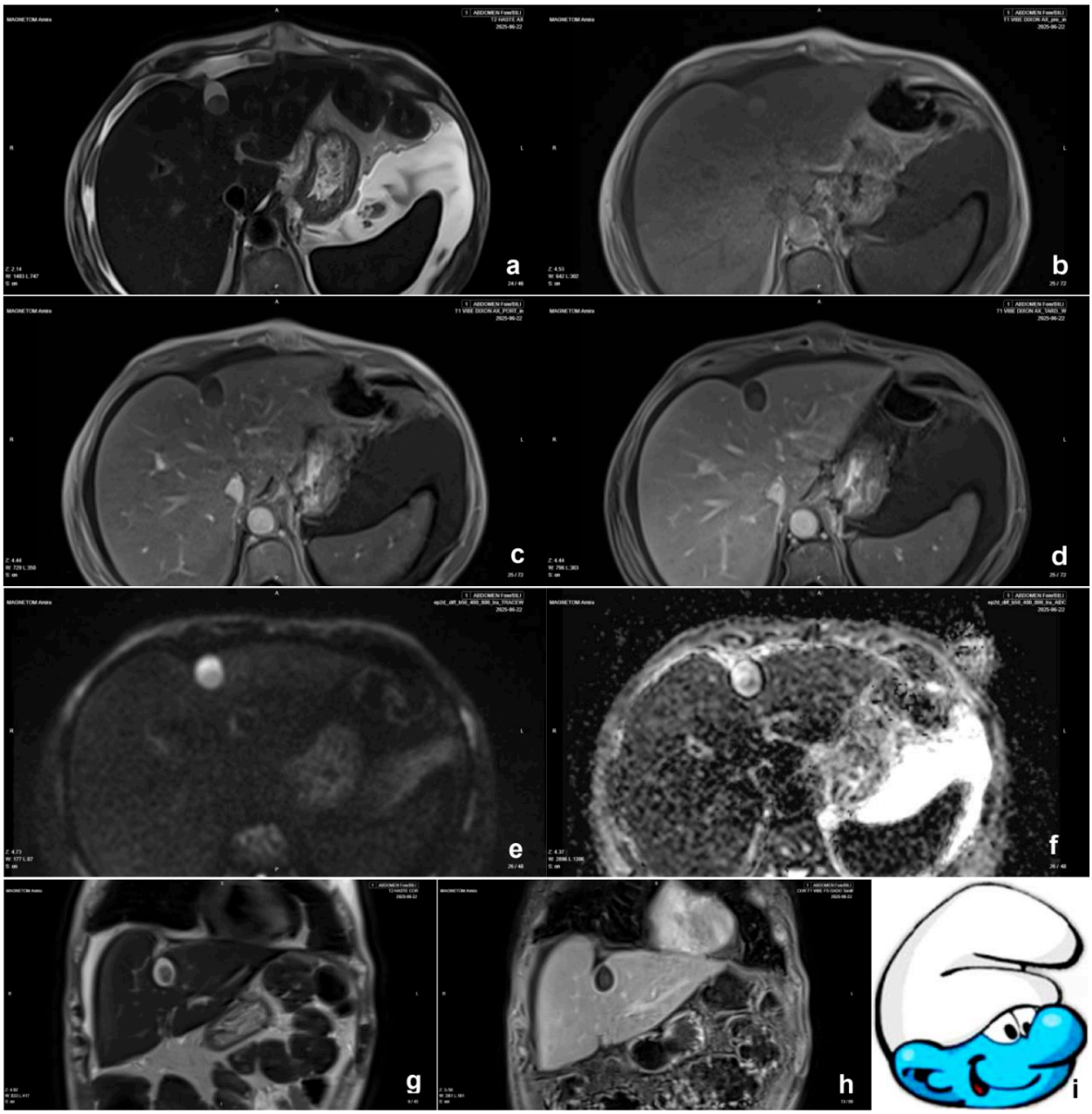

Figure 3. *Axial (a-f) and coronal (g, h) MRI images, (i) illustration[2] : T2-weighted (a, g), pre-contrast TI DIXON InPhase (b), post-contrast portal phase (c), delayed hepatic phase (d, h), diffusion-weighted imaging (e) with ADC map (f), showing a hyperintense T2 lesion (a, g), hypointense T1 (b) with dependent material of intermediate T2 signal (a, g) and hyperintense T1 (b), producing the "smurf's head" (i) appearance[2] on coronal images (g-h). No diffusion restriction (e, f). No enhancement of the cyst contents (c, d, h), but moderate delayed mural enhancement (d, h)*

images (4, 6, 11), without enhancement (4, 11) or diffusion restriction. Occasionally, iso- or hypointense T1 (10) signal may be seen, along with subtle delayed peripheral enhancement (6). The presence of dependent material with intermediate T2 and hyperintense T1 signal (6) produces the pathognomonic “smurf’s head” appearance (2), as seen in our case.

Once the diagnosis is established, histological confirmation is recommended, either by surgical excision or image-guided biopsy (12). Management remains controversial: both surgical resection and imaging surveillance are proposed (12), as CHFC is not purely benign due to its malignant transformation potential.

Conclusion

CHFC is a rare benign liver lesion that should be considered in any middle-aged adult presenting with a unilocular cystic lesion in segment IV. Given the risk of malignant transformation, surveillance or surgical excision should be discussed. MRI is the imaging modality of choice for diagnosis, allowing accurate lesion characterization.

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Declarations

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