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Ciliated Hepatic Foregut Cyst: A More Common Entity Than Previously Thought

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Abstract **Case Report**

Ciliated hepatic foregut cyst (CHFC) is a congenital cystic lesion of the liver derived from the embryonic foregut epithelium. Although usually asymptomatic, some present with non-specific abdominal symptoms. CHFC should be investigated thoroughly since they carry a risk of malignant transformation into squamous carcinoma, and to rule out other possible differential diagnoses. Owing to the variable imaging features, histological confirmation remains crucial. We present the case of a CHFC as well as a comprehensive literature review. Awareness of its imaging features is essential to include CHFC in the differential diagnosis and guide appropriate management.

Keywords: Ciliated Hepatic Foregut Cyst, Liver Cyst, MRI and CT Imaging.

INTRODUCTION

Ciliated hepatic foregut cyst (CHFC) is a typically benign hepatic lesion arising from the embryonic remnant of the foregut epithelium [1, 2]. Since its first description, only a limited number of cases have been reported in the literature, which explains why it remains unfamiliar to many radiologists and surgeons [1-3].

Most CHFC are asymptomatic and incidentally discovered during imaging modalities performed for unrelated complaints. However, they may occasionally present with non-specific abdominal pain or compressive symptoms, especially when the lesion is larger [2, 3].

Although the lesion is generally benign, its importance lies in two main issues : its ability to mimic several cystic hepatic lesions, and its potential for malignant transformation into squamous cell carcinoma on rare occasions [4, 5].

Imaging plays a central role in detection, initial characterization and follow-up of CHFC. They are particularly challenging due to their variable imaging appearance, which depends largely on cyst content (proteinaceous, mucinous, or fatty components) [4, 6].

Complete surgical resection is the primary therapeutic approach, with laparoscopy being an attractive option due to its postoperative and cosmetic advantages [7].

In this article, we describe the radiological appearance of a CHFC in a 64-year-old man, which was confirmed by histopathological examination, emphasizing the radiological features and diagnostic pitfalls.

CASE REPORT

A 64-year-old man presented with a two-month history of persistent right upper quadrant abdominal pain. He did not report fever, jaundice, weight loss or digestive disturbances. There was no known history of chronic liver disease, hydatid exposure or prior hepatobiliary surgery.

Physical examination was unremarkable. Laboratory investigations, including complete blood count, inflammatory markers, and liver function tests were within normal limits, showing no evidence of cholestasis or infection.

The patient underwent abdominal computed tomography (CT) as part of the workup, revealing an isodense hepatic lesion located in the anterior subcapsular region of segment IVa. No coarse calcifications or enhancement was observed after contrast administration. Its attenuation was slightly

higher than simple water density supporting the cystic nature of the lesion. (Figure 1).

Magnetic resonance imaging (MRI) serves as a sensitive modality and provided further characterization. It confirmed a well-defined lesion that appeared hypointense on T1-weighted images and markedly hyperintense on T2-weighted images. This lesion was the seat of a sediment creating a fluid-fluid level which was hyperintense on T1-weighted imaging and hypointense on T2-weighted imaging. No significant enhancement was identified after gadolinium administration, and no diffusion restriction was demonstrated. Also, no direct communication with the biliary ducts was detected. (Figure 2).

Given its location and the benign appearance on CT and MRI, diagnosis of CHFC was proposed and the decision was made to proceed with surgical resection.

Histopathological analysis confirmed the presence of a cyst lined by ciliated pseudostratified epithelial cells, consistent with CHFC.

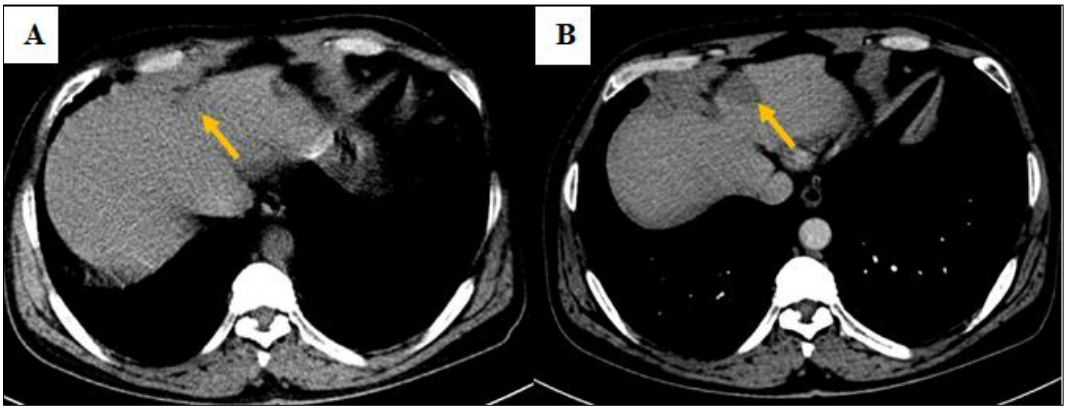

Figure 1: Non-contrast axial CT images (A) showing an isodense hepatic lesion located in the anterior subcapsular region of segment IVa. Post-contrast axial CT images (B) demonstrated no evident enhancement with an attenuation slightly higher than simple water.

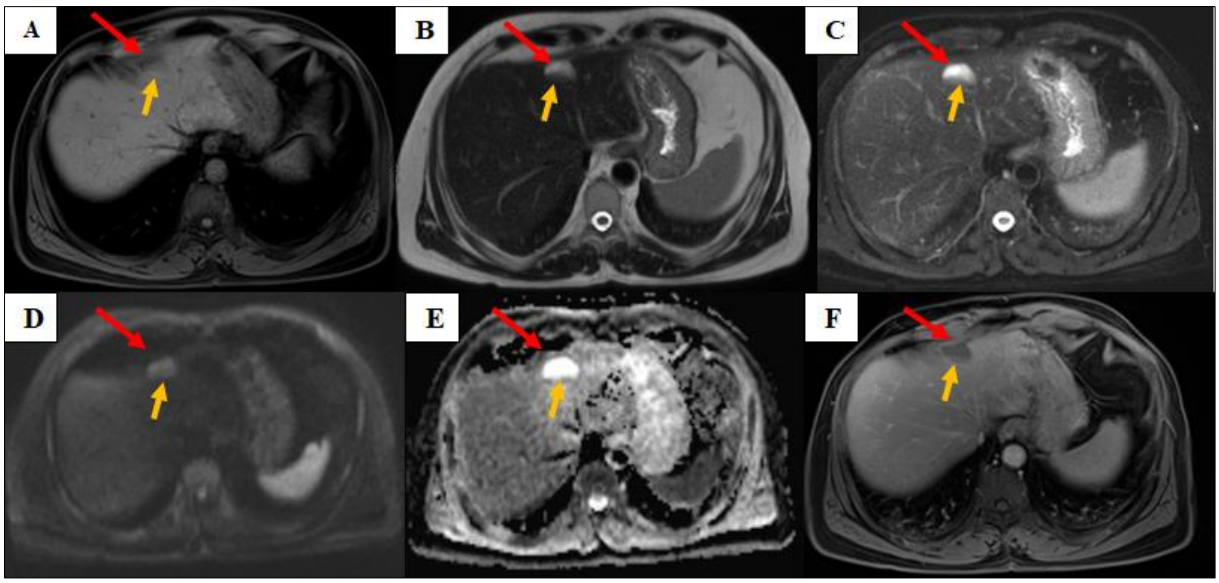

Figure 2: Axial MR images demonstrating a well-defined lesion (red arrows) hypointense on T1 (A), markedly hyperintense on T2 (B,C). It was the seat of a sediment creating a fluid-fluid level (yellow arrows) hyperintense on T1 (A) and hypointense on T2 (B,C). No diffusion restriction was demonstrated (D,E) and no significant enhancement was identified after gadolinium administration (F)

DISCUSSION

CHFC is a congenital lesion thought to arise from detached foregut epithelium that becomes incorporated into the liver during early embryonic development [1-4]. Histologically, it is characterized by four distinct layers including a pseudostratified ciliated columnar epithelium, subepithelial connective tissue, a smooth muscle layer, and an outer fibrous capsule [1-4].

An increasing number of CHFC are being reported in the literature, because of increased availability of radiologic imaging techniques [2-9].

From an epidemiological standpoint, they typically occur in adults with a median age between 50 and 55 years [10, 11], and are extremely rare in children [12].

They occur more frequently in men, most commonly located in segment IV of the liver and are usually subcapsular, unilocular, and relatively small [4-13]. However, atypical locations, including right lobe lesions and even extrahepatic locations such as the gallbladder, have also been described [6-14].

The clinical presentation varies, most cases in the literature were diagnosed incidentally or during surgery. However, a few reported cases presented with right upper quadrant abdominal pain as reported in our case, jaundice, and rarely portal hypertension, which was caused by mass effect by the cyst to adjacent tissues [2-11].

The radiological appearance of CHFC is variable, often attributed to the elements of the cyst contents [4].

On ultrasound, it is usually described as a well-defined hypoechoic or anechoic lesion, sometimes with internal echoes or dependent hyperdense sediment [1-3].

On CT, it generally appears as a spontaneously hypodense lesion, but its density may be slightly higher than water or even hyperdense depending on cyst content [10]. This CT variability is one of the main reasons why CHFC may be confused with hemorrhagic cysts, infected cysts, or cystic neoplasms [1-3].

MRI is the most informative and key imaging modality. Most CHFC are hyperintense on T2-weighted images, although usually not as strongly hyperintense as a simple cyst [4]. T1 appearance is more heterogeneous and may vary from low to high signal, particularly when the cyst contains proteinaceous, mucinous, or fatty material [1-15]. The presence of a fluid-fluid level or dependent T1 hyperintense sediment has been specifically emphasized in imaging series and case reports such as ours [3, 4]. This feature may be helpful, but it is not exclusive to CHFC.

An important point in the imaging assessment is the absence of enhancement after contrast administration [3, 4]. This helps distinguish CHFC from mucinous cystic neoplasms or biliary cystadenomas, which are more likely to demonstrate thick septations, mural nodularity, or enhancing walls [1]. Likewise, lack of diffusion restriction favors a benign cystic lesion rather than an abscess or highly cellular neoplasm [4].

Other differential diagnosis, either benign or malign, may include simple hepatic cyst, hydatid cyst, epidermoid cyst, intra-hepatic choledochal cyst, mesenchymal hamartoma and cystic metastasis [9-11]. An integrated approach combining clinical history, biological findings, and imaging characteristics is essential to narrow down the differential diagnosis [3].

Besides the diagnostic challenges that CHFC imposes, the management remains controversial [6-10]. The literature strongly supports complete surgical excision instead of long-term follow-up in symptomatic patients or in cysts with worrying imaging features [1-4]. This approach is justified by the rare but real risk of malignant transformation, generally into squamous cell carcinoma [8-17]. Reported malignant cases have typically involved larger lesions, wall abnormalities, thick septations and rapid growth as risk factors. Some authors recommend surveillance for small and asymptomatic lesions, but no universally accepted follow-up protocol exists [1-4].

CONCLUSION

Ciliated hepatic foregut cyst is is an increasingly frequently diagnosed condition, that should be included in the differential diagnosis of cystic liver masses. Imaging is essential for detection and characterization, but it is not sufficiently specific to establish a definitive diagnosis in many cases. Thus, histopathological confirmation remains crucial.

This report highlights the importance of considering this more common pathology in the differential diagnosis of hepatic cystic lesions, and underscores the need for surgical management given the potential risk of malignant transformation.

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