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Case Report

Ciliated hepatic foregut cyst: a rare diagnosis behind a hepatic cyst

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ABSTRACT

Ciliated hepatic foregut cysts (CHFC) are rare and typically benign lesions of the liver derived from an embryonic remnant of foregut epithelium. Patients are almost always asymptomatic and mostly incidentally found on radiologic studies. Radiographic imaging is usually insufficient for diagnosis and definitive diagnosis is made through histology. It is important to consider this condition in the differential diagnosis of liver lesions, particularly those located in the subcapsular region of the segment 4. Usual diagnostic uncertainty and a malignant potential that cannot be totally excluded, surgical excision should be considered. We describe a case of a 76-year-old male with an atypical cystic mass in segment 4 of the liver who underwent a laparoscopic resection. Histology revealed a ciliated hepatic foregut cyst.

Keywords: Ciliated hepatic foregut cyst, Liver cyst, Laparoscopic resection, Foregut malformation, Case report

INTRODUCTION

Ciliated hepatic foregut cysts (CHFC) are rare, typically benign, congenital hepatic cystic lesions of embryological origin that have been increasingly diagnosed in the last few decades with the increasing use of high-sensitivity abdominal imaging.[1]

This condition was first described in 1857, by Friedreich.[2] Wheeler and Edmondson in 1984 were the first to use the term CHFC and showed their histological similarities to bronchogenic and oesophageal cysts derived from the foregut.[3] It was the presence of shared histological features in the form of pseudostratified ciliated columnar epithelium that led them to describe these cysts as an anomaly resulting of evagination of the foregut during embryogenesis. Histologically, CHFCs classically consist of 4 layers, including a ciliated pseudostratified columnar epithelium with interspersed mucus cells, subepithelial connective tissue, a smooth muscle layer, and an outer fibrous capsule.[4]

We report a case of a patient who underwent successful laparoscopic resection of a CHFC. The clinical characteristics, diagnosis and treatment of these lesions are further discussed.

CASE REPORT

A 76-year-old caucasian male, with no relevant medical history, was referred to our hospital for further evaluation of a hepatic lesion incidentally found on abdominal ultrasound. His chief complaint was occasional right upper quadrant pain, and apart from minimal right upper quadrant tenderness examination was unremarkable.

Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a unilocular, subcapsular, cystic

mass with atypical features, located in segment 4b of the liver and measuring 4 cm. A CT scan of the abdomen showed a hypodense lesion that did not enhance after contrast injection and the MRI showed a hypointense lesion on the T1 spin echo and strongly hyperintense features on the T2 images.

The serological tumour markers including alpha-fetoprotein (AFP), carcinoembryonic antigen (CEA), carbohydrate antigen (CA) 19-9, and the rest of the biochemical profile were normal. Serological tests for hydatid cyst were negative.

In view of the diagnostic uncertainty, the cyst was excised. A laparoscopic hepatectomy of the mass in the left lobe was performed. During the surgical procedure, we found a unilocular, well-circumscribed and protruding cystic mass in the segment 4b of the left lobe, with a thick and intact cyst wall, consistent with prior imaging. Intraoperative ultrasound was performed and there was no need to apply vascular control. We removed the cystic mass completely along with liver tissue 5 mm away from the lesion, with no violation of cyst integrity or potential intraperitoneal spillage of contents.

The surgery and postoperative period were uneventful, and the patient was discharged on postoperative day 1.

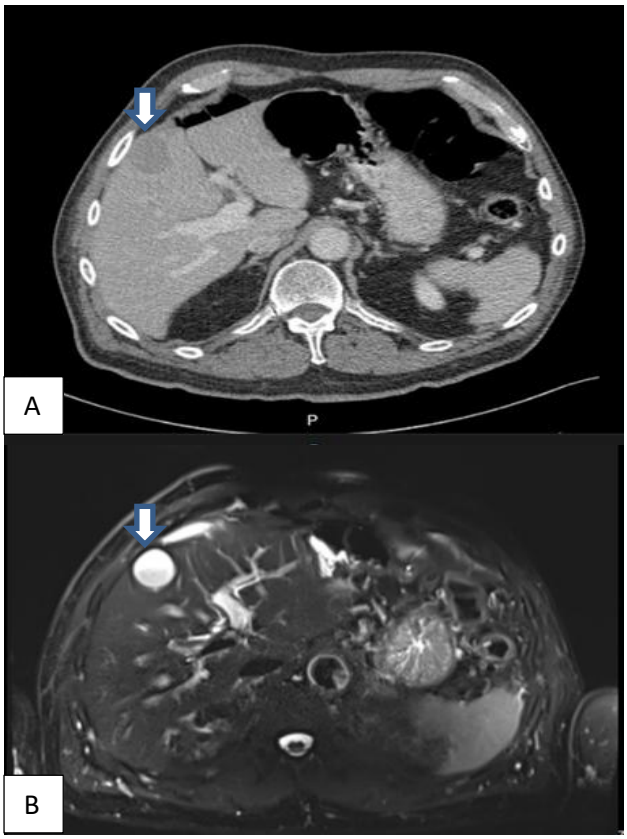

Figure 1 (A and B): Contrast-enhanced CT of the abdomen shows a hypodense lesion in segment 4; B) In MRI, the lesion exhibits high intensity in T2-weight images.

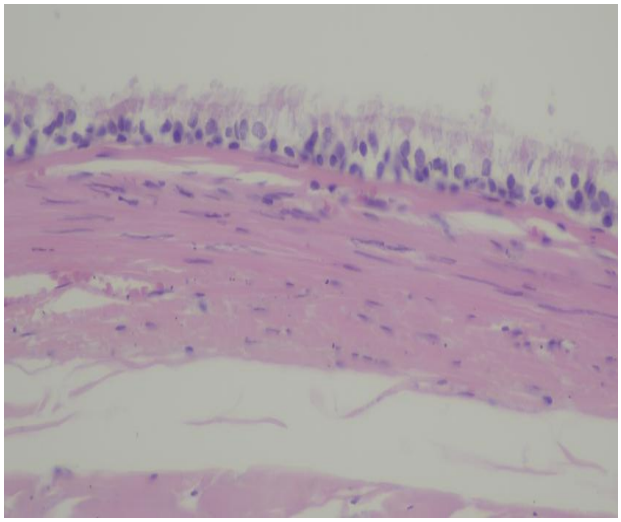

Figure 2: High power view of the cyst wall showing the pseudostratified ciliated epithelium, subepithelial connective tissue, smooth muscle layer and outer fibrous capsule (hematoxylin-eosin staining, 400× magnification).

The surgically removed cystic lesion was 3.9 cm in greatest diameter with a tan-pink capsule containing yellow mucinous fluid and a smooth whitish lining. Microscopic examination showed ciliated columnar epithelium, with subepithelial connective tissue stroma and a smooth double muscle layer. Immunohistochemistry showed the ciliated cyst lining positive for TTF1 and negative for MUC 5 AC and MUC2 with absence of expression of CD10. There was no evidence of malignancy. Based on these results a diagnosis of CHFC was made. At 5 years of follow-up, the patient has done well without postoperative complications or recurrence of the mass.

DISCUSSION

An early review summarised 103 cases in the literature; more recent reviews estimate that over 200 cases have now been reported.[4,17] CHFCs occur more frequently in men and are usually discovered during the fifth decade of life, but may present at any age.[1,5] Clinically, most patients are asymptomatic at the time of diagnosis and the cyst is found incidentally by abdominal imaging techniques, surgical exploration or at autopsy. When symptomatic, some patients may present with right upper quadrant abdominal pain, jaundice from biliary obstruction or portal hypertension.[1,6]

The shape and the anatomic location of the lesion are also important diagnostic considerations. CHFCs are most often solitary, subcapsular, unilocular, small (<4 cm) and fluid-filled but may present with findings suggestive of solid debris.[7,8] Classically, CHFCs are in the left lobe of the liver, especially segment 4, but can present within the right lobe as well.[1] The common location in segment 4 of the liver may be explained by the fact that this segment

and in particular the left lobe of the liver constitutes the bulk of the entire liver during the fourth to sixth weeks of development.[1] Furthermore, extrahepatic locations, such as the gallbladder and extrahepatic biliary system, have been reported.[9-11]

Although these cysts have suggestive features on imaging, radiology is insufficient for definitive diagnosis. The inner epithelial layer produces a thick mucoid content resulting in an increased density on radiologic imaging that can give the impression of a solid tumour. Ultrasound images usually reveal a round shape, a smooth wall and hypoechoic or anechoic cyst.[8] Another common characteristic on ultrasonography is the development of a complex cyst with echogenic material in its lumen.[12] A CT scan reveals a round lesion with variable density that reflects the variable fluid viscosities, and a feature that is commonly noticed is the lack of enhancement on contrast injection.[6,8] Large variability is seen on MRI T1-weighted imaging, from mildly hypointense to hyperintense, with the sign intensity probably reflecting the fluid viscosity, however, CHFCs are nearly exclusively hyperintense on T2-weighted imaging.[7,8] Caution should be exercised with respect to bleeding of the cyst, which significantly affects intensity and gives rise to hypointense regions in T2-weighted images.[6]

The differential diagnostic possibilities include simple cysts, mucinous cystic neoplasm, pyogenic abscess, amoebic abscess, hydatid cyst, intrahepatic pseudocyst, biliary cystadenoma, cystadenocarcinoma and cystic metastatic tumour.[5]

Definitive diagnosis is made through histology, and fine needle aspiration cytology may provide a nonsurgical diagnosis with reports of a positive predictive value of 76%.[1,5] The finding of ciliated pseudostratified tall columnar epithelial cells in a mucoid background is nearly diagnostic, as no other liver pathology presents with similar cytology.[5]

Although CHFCs were originally believed to be benign non-neoplasic lesions, recent reports of CHFCs undergoing malignant transformation have been reported.[13,14] Rates of malignant transformation were reported to range from 3% to 5%, with squamous cell carcinoma being the most common malignancy.[1,5] The main risk factor for malignant transformation in CHFCs appears to be its size.[1,5] Rates of malignant transformation are lower than those seen in mucinous cystic neoplasm, which may be as high as 10%.[5] The role of CA 19-9, and/or CEA as a marker of malignant progression in CHFC does not appear useful. There have been reports of patients with CHFC discovered to have elevated serum CEA, and elevated intra-cystic CEA and CA 19-9 in the absence of malignancy.[11,13]

The current management of these lesions remains controversial.[18.19] As a result of the potential risk of malignant transformation, most agree that surgical resection, increasingly performed through a laparoscopic approach, should be performed if the cyst is greater than 4-5 cm, symptomatic, growing, or has a mass formation in the cyst wall on imaging.[15,16] In the absence of concerning radiographic features for malignancy, a more conservative approach with serial imaging could be considered given the overall low risk of malignant transformation, particularly when lacking the above features.[5]

CONCLUSION

CHFCs are congenital cystic lesions that arise from the embryonic primitive foregut. It is difficult to determine the true prevalence of CHFCs because they are generally asymptomatic. Increased use of abdominal imaging techniques has improved detection rates of this lesion, but they remain extremely rare, and several crucial characteristics remain poorly understood, including the risk of malignant conversion. Diagnostic doubts are frequent when in the presence of a cyst hepatic lesion with atypical radiographic signs like CHFC, therefore, it is an important diagnosis to consider in the setting of a hepatic lesion and, as malignant potential cannot be totally excluded, surgical excision should be considered.

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