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Case Report

Xanthogranulomatous cholecystitis on fusion of the planes of the liver ☆

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ABSTRACT

Xanthogranulomatous cholecystitis (CXG) is a rare entity of cholecystitis, characterized by the presence of xanthogranulomas within the gallbladder wall, that could be misdiagnosed as a vesicular carcinoma. We report a case of 66-year-old man with xanthogranulomatous cholecystitis associated with an incidental finding of a fusion of the planes of the liver which is a rare anatomic variant. Imaging especially ultrasounds, CT scan, and MRI play a key role in the characterization of those anomaly, thus avoiding a non–suitable surgical procedure.

Introduction

Xanthogranulomatous cholecystitis (CXG) is a rare and uncommon form of chronic cholecystitis that could be misdiagnosed as vesicular carcinoma. Fusion of the planes of the liver is a rare anatomic liver variant characterized by the absence of the hepatic segment IV. We herein report a case associating these 2 rare entities.

Case presentation

A 66 years old man without any pathologic history, was admitted in the emergency department for right hypochondrium pain evolving for 2 months. The clinical examination found a sensitivity of the right hypochondrium with no other abnormality.

☆ Competing Interests: We testify that the content of the manuscript is original We confirm that this manuscript has not been published elsewhere and is not under consideration by another journal. There are no conflicts of interest to disclose. Thank you for your consideration of this manuscript.

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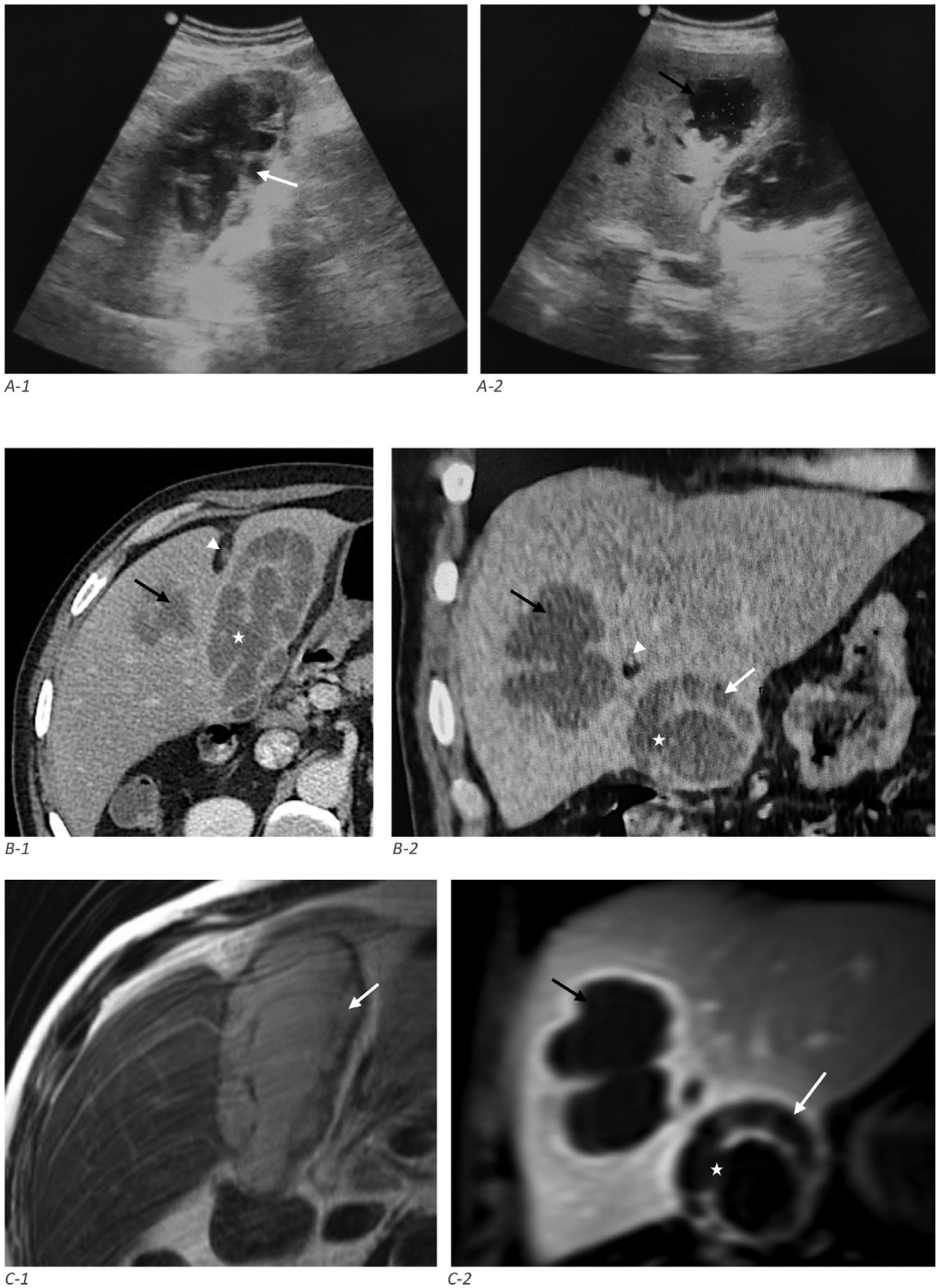

Fig. 1 – Perforated xanthogranulomatous cholecystitis associated with abscess and fusion of the hepatic planes. (A) Abdominal ultrasound: Transverse section through the gallbladder (A-1) and liver collection (A-1); (B) Abdominal CT scan with injection: axial section (B-1) and coronal reconstruction (B-2); (C) sequence MRI with T2 weighting in the axial plane (C-1) and T1 weighting in the coronal plane (C-2). Showing multiple parietal nodules (black arrow): hypoechoic in ultrasound, hypodense CT and hyposignal T1, intermediate T2 signal in a diffusely thickened vesicular wall measuring 14 mm. An area of mucosal defect (white star) with associated intramural collection and an adjacent liver collection (white arrow). The round ligament is aligned with the gallbladder in the same plane, suggesting fusion of the hepatic planes (white arrowheads).

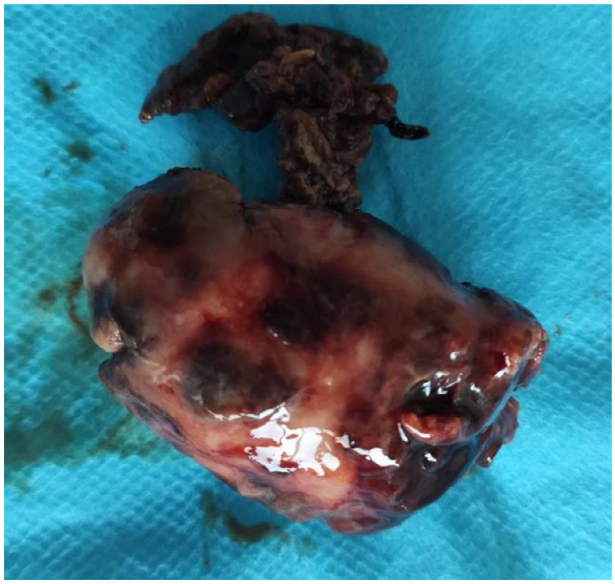

Fig. 2 – Cholecystectomy specimen showing a pseudotumoral aspect of the gallbladder.

The biological assessment showed hyperleukocytosis at 17,000/uL (normal: 4000-10,000), an elevated C- reactive protein at 215 mg/L (normal <5 mg/L), and a biological cholestasis.

Imaging showed a diffusely thickened gallbladder wall measuring 14 mm with multiple nodules within. Also described an area of mucosal defect with associated intramural collection and an adjacent liver collection. We incidentally noted a round ligament aligned with the gallbladder in the same plane, suggesting a fusion of the median planes of the liver.

Those specific findings associated with the absence of dilatation of the intrahepatic bile ducts and the absence of hepatic invasion suggested the diagnosis of CXG (Fig. 1).

A cholecystectomy with drainage of the hepatic abscess was performed subcostally (Fig. 2). Pathologic examination confirmed the diagnosis of CXG and the evolution was without any complication.

Discussion

CXG is a rare form of chronic cholecystitis. Its incidence is highest in the sixth and seventh decades of life. The association with vesicular lithiasis is found in 97% of cases [1], while no association with fused liver plans has been reported in the literature. Its pathophysiology is poorly understood and is probably secondary to rupture of the Rokitansky-Aschoff sinus or to mucosal ulcerations in the gallbladder wall responsible for parietal biliary infiltration [2]. This intramural bile is degraded by histiocytes leading to the formation of xanthomatous cells (foamy histiocytes), a chronic granulomatous inflammatory response, and finally a fibrous reaction. Clinically, CXG manifests itself as acute or chronic cholecystitis. Examination may find a palpable mass or a positive Murphy's sign. The biological assessment often finds hyperleukocytosis and a disturbance of the liver test with cytolysis or cholestasis. Elevation of the tumour marker CA19-9 is common in the CXG and therefore cannot be used for the differential diagnosis of vesicular carcinoma [3].

In ultrasonography, the appearance may be characteristic with a diffuse thickening of the gallbladder wall, often more than 10 mm, which is the site of hypoechoic nodules. This characteristic appearance is found in only half of the cases [4]. The nodules may appear iso- or hyperechoic, or may be absent. The CT scan shows a thickening of the gallbladder wall, which is diffuse in 91% of cases [5]. The thickened wall is the site of hypodense nodules corresponding to xanthogranulomas or sometimes abscesses. Xanthogranulomas can be superinfected, leading to the formation of parietal abscesses with a risk of perforation, and fistula. Usually, the mucosal surface remains intact contrary vesicular carcinoma. The visibility of a continuous mucosal line that increases after injection is in favor of CXG [6]. CT scan may reveal periventricular infiltration extending to the liver, duodenum or colon which may lead to misdiagnosis. Regional lymph nodes can be found, but are more common in vesicular carcinoma [7]. In the majority of cases, MRI shows a parietal thickening as a heterogeneous hyposignal in T1, a hypersignal in T2, with a drop in signal in phase opposition in relation to the presence of intramural fat [8]. In our observation a fusion of the median planes of the liver was identified. In this rare anatomic variant, segment IV and its portal branch do not exist, with a variation in portal division. It is probably the consequence of a lack of embryonic development of segment IV. The gallbladder then lies in the same plane as the round ligament [9].

PET SCAN is rarely indicated, with a high frequency of false positives showing hyperfixation even in the absence of malignancy [10]. In patients suspected of malignancy, an ultrasound-guided percutaneous or endoscopic puncture biopsy can be performed [11] to avoid iatrogenic complications. The treatment is essentially surgical, consisting of a cholecystectomy. Pathologic examination confirms the diagnosis of CXG by the presence of foamy histiocytes, inflammatory infiltrate, and parietal fibrosis.

Conclusion

CXG is a rare form of chronic cholecystitis that can be mistaken for vesicular carcinoma, which is difficult to diagnose preoperatively. Imaging can sometimes be used to determine the characteristic appearance of the disease, thus avoiding a disproportionate surgical procedure. The fusion of the median planes of the liver is a rare anatomic variant which should be pointed out, as it carries a risk of liver surgery complications when it is unknown.

Patient consent

Informed consent was obtained from the patient for the publication of this case report.

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