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Xanthogranulomatous Cholecystitis is not always a Cancer

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Abstract | **Case Report**

***Introduction*:** Xanthogranulomatous cholecystitis is a rare condition with a specific symptoms. The main differential diagnosis is vesicular adenocarcinoma. It can be revealed by abdominal pain, hemorrhage or fistulas. We report here the case of a patient with xanthogranulomatous cholecystitis revealed by febrile abdominal pain. We report a case of xanthogranulomatous cholecystitis in a 45 year old woman, revealed by febrile abdominal pain. ***Case Report*:** The patient was 45 years old and had no particular pathological history. She presented with acute febrile abdominal pain. The clinical examination revealed a conscious patient, hemodynamically and respiratorily stable, febrile (fever 39°C), with abdominal defence of the right hypochondrium, the rest of the examination was without anomaly. An abdominal CT scan was requested urgently revealing a circumferential and regular parietal thickening of the gallbladder, hypodense with spontaneous contrast enhanced after injection of PDC measuring 12 mm of maximum thickness, with individualization of multiple fissured calculi, seat of air bubbles realizing the sign of "Mercedes-Benz sign", It is associated with an important infiltration of the mesenteric fat of the neighborhood. A complementary MRI scan with T1, T2, Diffusion, 3DMRCP sequence was performed, showing a parietal thickening in T1 hyposignal, T2 intermediate signal, diffusion hypersignal, multiple liquid structures in T2 hypersignal associated with multiple stones in signal void on all sequences, and a dilatation of the main bile duct measuring 13 mm upstream of a regular eccentric stenosis of the lower bile duct with an inflammatory appearance. ***Discussion and Conclusion*:** Xanthogranulomatous cholecystitis is a pathology that is difficult to diagnose preoperatively, as it can be mistaken for a vesicular adenocarcinoma. CT and MRI scans can sometimes help to clarify the diagnosis, particularly in the presence of a diffuse and continuous thickening of the wall with a continuous mucous line, hypodense intramural nodules, and the presence of lithiasis.

Keywords: Xanthogranulomatous cholecystitis, vesicular adenocarcinoma, hypochondrium, Mercedes-Benz sign.

INTRODUCTION

Xanthogranulomatous cholecystitis (CXG) represents a rare histological subtype of cholecystitis, first described in 1970 by Christensen and Ishak as a "benign pseudotumor of the gallbladder". It is rarer than gallbladder carcinoma (GBC), which is frequently diagnosed as a first-line disease. This benign pathology is particularly important to recognize, because the extension of these lesions beyond the limits of the gallbladder compartment exposes, in case of confusion with CVB, to inappropriate and disabling surgery. Furthermore, the association of CXG and CVB remains a matter of debate in the literature. Because of these diagnostic uncertainties, the management of this entity is complex and unsystematized.

We report a case of xanthogranulomatous cholecystitis in a 45-year-old woman, revealed by febrile abdominal pain.

CASE REPORT

The patient was 45 years old, without any particular pathological history, and presented with acute febrile abdominal pain. The clinical examination revealed a conscious patient, hemodynamically and respiratorily stable, febrile (fever 39°C), with abdominal defence of the right hypochondrium, the rest of the examination was without anomaly.

An abdominal CT scan was requested urgently revealing a circumferential and regular parietal

thickening of the gallbladder, hypodense with spontaneous contrast enhanced after injection of PDC measuring 12 mm of maximum thickness, with individualization of multiple fissured calculi, seat of air bubbles realizing the sign of "Mercedes-Benz sign", It is associated with an important infiltration of the mesenteric fat of the neighborhood.

A complementary Bili-MRI with T1, T2, Diffusion, 3DMRCP sequence was performed showing a parietal thickening in T1 hyposignal, T2 intermediate signal, diffusion hypersignal, multiple liquid structures in T2 hypersignal associated with multiple stones in signal void on all sequences, and a dilatation of the main biliary tract measuring 13 mm upstream of a regular eccentric stenosis of the lower bile duct of inflammatory appearance.

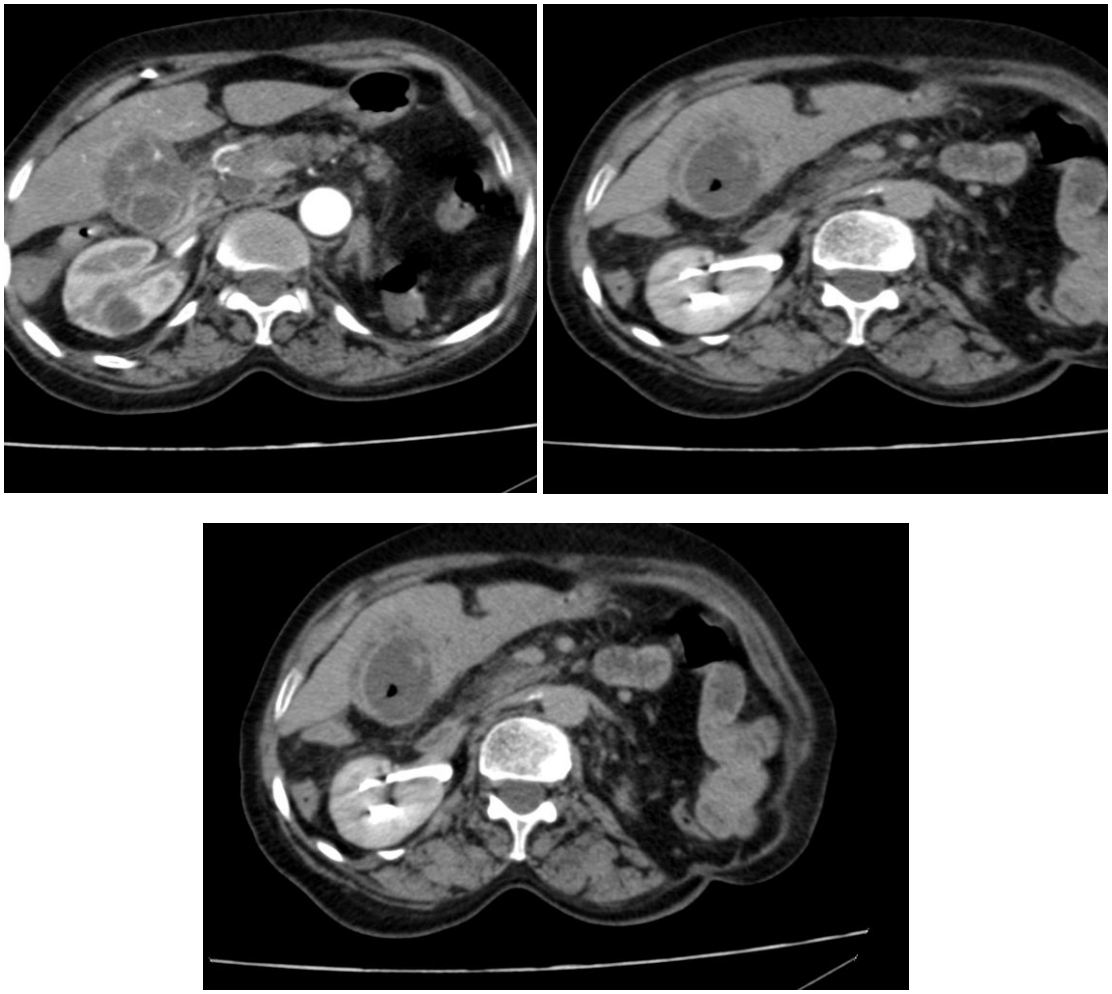
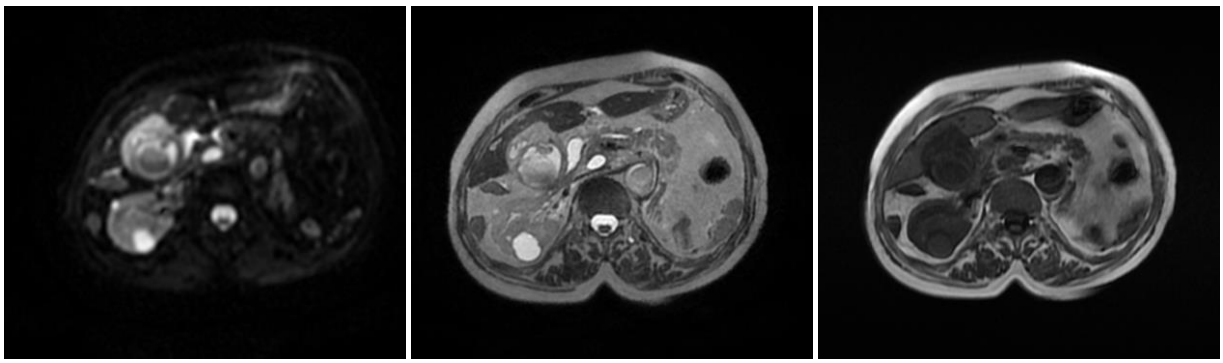


Figure 1: Abdominal CT scan in axial sections with and without injection: Circumferential and regular parietal thickening of the gallbladder hypodense with spontaneous contrast enhanced after injection of PDC measuring 12 mm of maximum thickness, with individualization of multiple fissured stones sitting on air bubbles realizing the "Mercedes-Benz sign". It is associated with an important infiltration of the neighbouring mesenteric fat



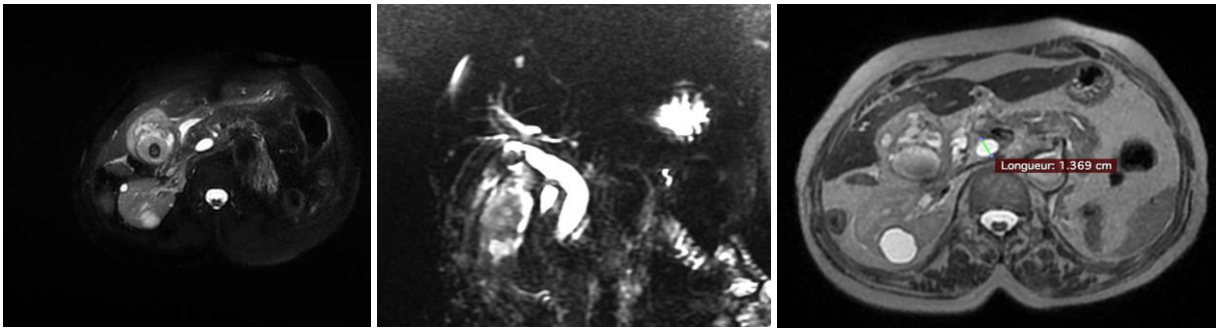


Figure 2: MRI scan with T1, T2, Diffusion, 3DMRCP sequence: T1 hyposignal parietal thickening, T2 intermediate signal, diffusion hypersignal, multiple T2 hypersignal fluid structures associated with multiple stones in signal void on all sequences. Dilatation of the main bile duct measuring 13 mm upstream of a regular eccentric stenosis of the lower bile duct with an inflammatory appearance

DISCUSSION

Xanthogranulomatous cholecystitis (CXG) is a rare entity of cholecystitis, characterized by the presence of prominent xanthogranulomas within the wall of a very often lithiasis gallbladder. Its pseudotumor presentation with sometimes invasion of the surrounding organs may mimic a cholangiocarcinoma of the gallbladder (CVB).

The clinical symptoms of CXG are usually nonspecific and similar to those of acute or chronic cholecystitis. Abdominal pain is predominant (84.6%), followed by nausea and vomiting (25.6%). Anorexia is observed in 18% of patients, associated with weight loss in 8.9% of cases. The latter is sometimes very important and particularly misleading in the differential diagnosis with CVB.

The most common associated clinical sign is a Murphy's sign (53%), sometimes with right hypochondrial curvature (9.5%) that may also mimic CVB.

CXG is often associated with an acute complication related to gallbladder stones: jaundice is present in 20.5% of patients, secondary to stone migration into the main bile duct, more rarely to Mirrizi syndrome. Acute biliary pancreatitis or gallbladder empyema may also occur concomitantly. Finally, extrinsic antropyloric compression is rare.

There is frequently an inflammatory syndrome with hyperleukocytosis and elevated CRP, associated with cytolysis, which is non-specific.Cholestasis with elevated conjugated bilirubin may be present, especially in cases of associated lithiasis. CA 19.9, measured in case of diagnostic doubt, may be elevated to several thousand during a CXG, but does not allow to conclude about the malignancy of the lesion. Normalization of CA 19.9 should be checked after treatment.

The challenge of imaging is to associate focal or diffuse thickening of the gallbladder with a CXB and to eliminate a CVB in order not to commit the patient to inappropriate therapeutic management. Whatever the imaging modality, the detection of intraparietal nodules (xanthogranulomas) is a determining semiological element.

Ultrasound is performed in the initial workup of a clinical picture suggestive of gallbladder disease. Gallstones (multiple, simple or sludge) are frequent but inconstant. There is a moderate or most often marked thickening of the vesicular wall, often diffuse and circumferential. The gallbladder is rather collapsed, but it can be distended, especially in case of a stone embedded in the cystic duct.

CT has moderate sensitivity (67-78%) and low specificity (22-33%) for distinguishing CXGs from CVBs, because of similar scanographic features. It can demonstrate the characteristic hypodense parietal nodules if they are diffuse, but in the presence of more focal involvement, they are more difficult to distinguish from perivesicular abscesses, or adenomyomatosis lesions, which may also present as hypodense elements of the vesicular wall. The continuous enhancement of the wall, with a late enhancement on its hepatic side, is a very suggestive feature, but a break in continuity may be observed in diffuse involvement, as well as in forms with intrahepatic extension. The CT scan shows very well the distant extension when it is present: extension to the liver parenchyma, to the duodenum, or even to the colon. It shows very well a possible arterial or portal venous invasion, or of the main biliary tract, which can impose for a CVB. Finally, it detects complications in the form of fistula or vesicular perforation.

Magnetic resonance imaging (MRI) has a better diagnostic performance than ultrasound and CT in this type of pathology, resulting in a sensitivity of 79-86% and a specificity of 94.7% in some studies. MRI objectifies this diffuse or focal thickening of the vesicular wall, in hypo- or iso-signal T1, enhancing late after injection. Parietal nodules, if small, may be less well seen than on CT, but the presence of fatty elements within these nodules or in the vesicular wall is a discriminating semiological element.

Its treatment is total or partial laparoscopic cholecystectomy, with a high risk of conversion and complication. In case of suspicion of xanthogranulomatous cholecystitis, a referral center in hepatobiliary pathology should be contacted.

CONCLUSION

CXG is a rare and benign form of cholecystitis, characterized histologically by xanthogranulomas in the vesicular wall. Its pseudotumor presentation may lead to the erroneous assumption of CVB. MRI is the most specific preoperative examination. Diffuse parietal thickening, hypovascular parietal wall nodules, and continuously enhancing mucosa are suggestive of CXG. The treatment is laparoscopic cholecystectomy, total or partial, with however a high risk of conversion and complication.

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