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CLINICAL CASES

Xanthogranulomatous cholecystitis. A rare case of chronic cholecystitis simulated gallbladder cancer

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Abstract

Introduction: *Xanthogranulomatous cholecystitis is a rare inflammatory disease of the gallbladder a variety of chronic cholecystitis. Some patients with this entity are misdiagnosed with gallbladder cancer, since the radiological findings and clinical manifestations are similar, which means unnecessary radical surgery, with increased morbidity and mortality.* **Case presentation:** *A 60-year-old male patient with a pre-operative diagnosis of cholecystitis plus dilation of the bile duct, and suspicion of a neoplastic process of the gallbladder in the post-operative period.* **Conclusion:** *The difficulty of an evident differential diagnosis after imaging techniques gives clinical relevance to the picture.*

Keywords: *Xanthogranulomatous cholecystitis. Gallbladder cancer. Chronic cholecystitis.*

Introduction

Xanthogranulomatous cholecystitis (CX) is a rare inflammatory entity of the gallbladder. CX is a variety of chronic cholecystitis with a very low frequency 0.7-13%[1,2] characterized by a destructive inflammatory process of a focal or diffuse nature, with an accumulation of layers of lipid macrophages, fibrous tissue, and acute and inflammatory cells. chronicles[3]. It is increasingly recognized by pathologists and clinicians, with an increase in its frequency identified. CX is more common than gallbladder cancer, in Japan it fluctuates from 0.7% to 1.8% of all cholecystectomy specimens, in the US from 1.2% to 10%, and 10% to 13.2% in India[4]. The occurrence in our environment of CX in surgically resected vesicles ranges from 1% to 2%.

CX was first described by Christensen and Ishak in 1970, as a pseudotumor of the gallbladder[5], but it was not until 1981 that Goodman and Ishak, from the Institute of Pathology of the United States Armed Forces, coined the term CX, due to its similarity to xanthogranulomatous pyelonephritis[6]. The diagnosis is histopathological; however, ultrasound and tomographic data that raise suspicion in this entity show the presence of gallstones (multiple or individual) most of the time[7]. Morbidity from this disease is 32% and no data are available regarding mortality[8].

The objective of the study was to describe the clinical, radiological, and surgical findings, as well as the histopathological results of a clinical case of a patient with CX diagnosed post-surgery as gallbladder cancer.

Case report

A 60-year-old male patient, with no pathological or surgical history, came to the emergency room due to abdominal pain of 2 days' duration in the epigastrium after eating; intensity of 8/10 on the pain scale, stabbing type that radiates to the lumbar region, associated with nausea without vomiting. The patient had scleral jaundice, abdomen with pain and resistance in the right

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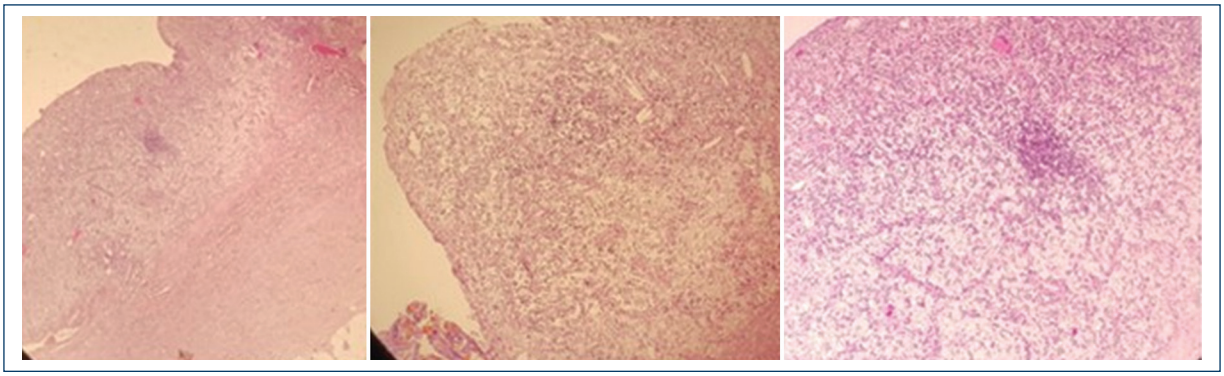

Figure 1. Histopathology of the gallbladder wall: typical macrophage histiocytes filled with debris and lipids; fibroblasts and accumulations of mononuclear lymphocytes with some giant cells.

upper quadrant, and a positive Murphy sign. The studies showed leukocytosis with left shift, preserved renal function, normal pancreatic enzymes, elevated bilirubin at the expense of direct bilirubin, and elevated liver enzymes with a cholestatic pattern. Abdominal ultrasound showed hepatomegaly with mild hepatic steatosis, dilation of the extrahepatic bile duct with a 9-mm common bile duct without evidence of an obstructive cause, gallbladder with WES complex, normal pancreas, without free fluid. Due to the worsening of the clinical picture and data of systemic inflammatory response, it was decided to take him to the operating room.

Open cholecystectomy shows a gallbladder with thickened hourglass walls with a stone fixed in the gallbladder fundus and in Hartman's pouch, abundant fibrosis containing thick bile, firmly adhered to the liver which made dissection of the bed difficult, Callot's triangle with marked fibrosis, thickened cystic duct with stenosis of its lumen, bile duct dilated approximately 10 mm, bile duct and intraoperative cholangiography without evidence of stones and adequate passage of distal and proximal contrast medium. The procedure is defined as extrinsic obstruction of the bile duct due to Mirizzi I Syndrome versus gallbladder neoplasia. The patient progressed adequately with progressive normalization of the liver profile and was discharged on the 4th day without complications. The pathological result was received with a CX report (Fig. 1).

Discussion

Various terms have been used to describe this process including ceroid granuloma, ceroid-type histiocytic granuloma, fibroxanthogranulomatous inflammation, and biliary granulomatous cholecystitis, although most authors now refer to it as CX[9]. CX is a pathology with a difficult diagnosis, frequently confused with gallbladder cancer; 40% of cases present intraoperative diagnostic doubts with this neoplasm. It is more common in women between 60 and 70 years old, generally with associated comorbidity, and in almost all cases, it is associated with long-standing cholelithiasis. Unlike the usual chronic cholecystitis, it is a process with a greater local infiltrative and destructive tendency[10].

Histologically, it is defined by the rupture of the Rokitansky-Aschoff diverticula, which are formed after a process of chronic cholecystitis, or by ulceration of the mucosa. This produces a severe inflammatory reaction in the interstitial tissue composed of fibroblasts and macrophages, which, in the process of inevitable phagocytosis of cholesterol and bile phospholipids, secondarily conditions a destructive enzymatic discharge of the tissue microenvironment of the gallbladder wall and its surroundings. This causes tumor-like thickening of the wall, with the appearance of yellowish nodules or striae that can extend to adjacent structures and organs with the formation of fistulas due to ulceration of the mucosa[11,12] and tissue fibrosis similar to the desmoplastic reaction typical of some tumors. epithelial, resulting in a macroscopic appearance similar to neoplastic[13]. Obstruction and chronic infection caused by the presence of stones produce degeneration and necrosis of the gallbladder wall, with the consequent formation of intramural abscesses. These are replaced by xanthogranulomas with exogenous foamy giant cell histiocytes, implicated as contributing factors similar to what has been experimentally demonstrated in xanthogranulomatous pyelonephritis[14].

Radiologically, CX can be confused with gallbladder cancer; one and the other can coexist in 10% of

cases[15]. Ultrasound shows hyperechogenic thickening of the gallbladder wall and the identification of intramural hypoechoic nodules representing foci of xanthogranulomatous inflammation or small abscesses, alteration of the mucosal lining, perivesicular fluid, stones, and bile sludge, and hyperechogenicity of the adjacent liver parenchyma[16,17]. Computed tomography shows, in addition to mural thickening, hypodense intramural nodules (5-20 mm), heterogeneous wall enhancement, and infiltration into the perivascular fat and adjacent liver parenchyma. Magnetic resonance imaging (MRI) shows xanthogranulomas with higher signal intensity on T2-weighted images. In-phase and out-of-phase sequences help identify intramural fat and diffusion sequences help differentiate CX from wall thickening due to gallbladder carcinoma[18]. In our case, MRI was not performed since it is not a routine diagnostic method in chronic cholecystitis.

Clinical symptoms, physical examination, and laboratory results are not useful to differentiate this pathology from other more common gallbladder or bile duct diseases. The clinical presentation does not differ from the spectrum of cholelithiasis and there are no specific characteristics of the disease. There are reports that describe the presence of a palpable mass in slightly less than half of the cases as well as mild jaundice in half of the patients who presented acutely. Some patients present perivascular or hepatic abscesses, gallbladder perforation, or enterobiliary fistulas, with figures ranging from 23% to 32%[19]. In our case, we found vesicle adhesions to loops of the small intestine, Mirizzi Syndrome I, and a decrease in the lumen of the cystic duct.

At present, cholecystectomy is preferable to initial medical treatment in acute cholecystitis; in the case of cancer, it should be performed by a surgeon with extensive experience in liver surgery[20]. When gallbladder cancer is suspected, it must be assumed that most cases are unresectable or have a poor prognosis and that laparoscopic surgery is contraindicated due to the increased risk of bile effusion, gallbladder rupture, peritoneal dissemination, and recurrence. Surgical management of gallbladder cancer is indicated in the earliest stages of the disease and it is important to evaluate therapeutic options in patients with advanced disease[21]. An expert consensus and the National Comprehensive Cancer Network guidelines recommend simple cholecystectomy in patients with T1a. For non-metastatic higher stages, extended oncological resection is recommended in patients with stages T1b, T2, and T3 who do not have disseminated disease. This surgery includes gallbladder fossa resection, IVb and V bisegmetectomy, and regional lymph node dissection; common bile duct resection and hepatectomies are considered in some selected patients. Patients with advanced stages (T4) treatment are palliative[22].

Many experts emphasize the need for general surgeons to become familiar with this pathology since CX can mimic hepatobiliary neoplasia in its clinical presentation, radiological findings, intraoperative appearance, and rarely, histological appearance, which often makes pre-operative distinction between these entities impossible[23].

Conclusions

CX is a rare pathology that usually affects patients with long-standing cholelithiasis. As it occurs in patients with greater comorbidity, post-operative complications also increase. CX can simulate a neoplasm or coexist with gallbladder carcinoma. Differentiation between CX and gallbladder cancer can be made intraoperatively by fine-needle aspiration biopsy or cold section. Due to the possible association between these two pathologies, the surgical procedure of choice should be total cholecystectomy. At present, it is impossible to macroscopically differentiate CX and gallbladder adenocarcinoma, so the definitive diagnosis is obtained until the histopathological analysis of the surgical specimen. CX can be a diagnostic challenge and a therapeutic dilemma due to its similarity to gallbladder neoplasia.

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Conflicts of interest

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Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

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