**Case Report**

**Xanthogranulomatous Cholecystitis Mimicking Gallbladder Adenocarcinoma: Case Report**

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**ABSTRACT**

Xanthogranulomatous cholecystitis is a rare disease characterized by severe fibrosis that can be a cause of technical difficulty in performing cholecystectomy (either laparoscopic or open surgery). Patients with xanthogranulomatous cholecystitis can be frequently confounded at intraoperative evaluation with gallbladder carcinoma, this fact can lead to an extended resection. The authors report one case of xanthogranulomatous cholecystitis that mimicked gallbladder adenocarcinoma at intraoperative evaluation by frozen-section. The patient was submitted to an extended resection (bisegmentectomy IV+V) with hilar lymphadenectomy. He carried out without postoperative complications. Definitive histological analysis revealed xanthogranulomatous cholecystitis.

**Key words.** Xanthogranulomatous cholecystitis; hepatectomy; laparoscopy; biliary neoplasms; surgery

**RESUMO**

COLECISTITE XANTHOGRAULOMATOSA SIMULANDO ADENOCARCINOMA DE VESÍCULA BILIAR: RELATO DE CASO

Uma colecistite xantogranulomatosa é uma doença rara caracterizada por intensa fibrose que pode ser uma causa de dificuldade técnica na realização de colecistectomia tanto por cirurgia laparoscópica quanto aberta. Casos de colecistite xantogranulomatosa podem ser frequentemente confundidos com carcinoma de vesícula biliar localmente avançado na avaliação intra-operatória fato esse que pode acarretar em uma ressecção alargada. Os autores relatam um caso de paciente com colecistite xantogranulomatosa que simulou adenocarcinoma de vesícula biliar localmente avançado tanto a avaliação intraoperatoriária quanto a biópsia de congelação. O doente foi submetido a ressecção estendida (bissegmentectomia IV+V) com linfadenectomia hilar e não houve intercorrências. À análise histológica definitiva, observou-se tratar de colecistite xantogranulomatosa.

**Palavras-chave.** Colecistite xantogranulomatosa; hepatectomia; laparoscopia; neoplasia de via biliar; cirurgia.

**INTRODUCTION**

Xanthogranulomatous cholecystitis is an idiopathic, very rare disease characterized by lipid-laden histiocytes deposits in gallbladder wall. It was first reported by McCoy in 1976 and described as distinct pathological condition by Goodman and Ishak in 1981. Xanthogranulomatous cholecystitis is a chronic inflammatory disease of gallbladder. The main characteristic is a bulky thickening of the gallbladder wall with a great propensity to infiltrate to adjacent organs, even leading to fistula formation.

Xanthogranulomatous cholecystitis generally presents as chronic or acute cholecystitis, and either findings from image examinations or even from intraoperative aspects may be mistaken for gallbladder carcinoma.

The objective of this report was to describe one case of xanthogranulomatous cholecystitis that mimicked gallbladder adenocarcinoma at intraoperative evaluation by frozen-section.

**CASE REPORT**

The case presented by the authors involved a 72-year-old white male who had no comorbidity factor. His clinical presentation was a three month biliary colic. Preoperative abdominal ultrasonography...
evaluation revealed multiple gallstones with thickened gallbladder wall. The patient underwent laparoscopic cholecystectomy and intraoperative findings included a hardened gallbladder, diffuse wall thickening and markedly adhesion to the liver without a surgical cleavage plane (Figure 1). Patient was converted to an open surgery because of the clinical suspect of gallbladder carcinoma. A hepatic resection of the segments IVB and V (inferior central hepatic bissegmentectomy) was performed in monobloc pattern (Figures 1 and 2). Frozen-section investigation was realized. Subsequently, gallbladder carcinoma was confirmed. Therefore, additional hilar lymphadenectomy was performed. Postoperative course was uncomplicated. Patient was discharged home on seventh postoperative day. The final diagnosis by histological examination revealed xanthogranulomatous cholecystitis.

Figure 1. Segments IV and V hepatic bissegmentectomy showing a tumoral gallbladder aspect with diffuse wall thickening and markedly adhesion to the neighboring liver without a surgical cleavage plane.

Figure 2. Intraoperative aspect showing inferior central hepatic bissegmentectomy (segments IVB and V) performed.

DISCUSSION
Xanthogranulomatous cholecystitis is an unusual presentation of chronic cholecystitis and often mimics a gallbladder carcinoma. Reports of its incidence range from 0.7% to 13.2% and it seem to become more prevalent after the age of 50-60 years.8 Gallstones have been described in the majority of xanthogranulomatous cholecystitis patients (92% to 100%).8 Factors as obesity and diabetes mellitus have been also frequently reported in these patients.8

Xanthogranulomatous cholecystitis may be associated simultaneously with a gallbladder cancer in 0.2% up to 12.5% cases.9 The premalignant nature of xanthogranulomatous cholecystitis is not known, however a recent study do not support a neoplastic condition of xanthogranulomatous cholecystitis, still indicating the inflammatory nature of the lesion.10

Diagnosis is a great challenge, preoperatively and even intraoperatively. Definite diagnosis is based on postoperative histopathological examination.

Clinical symptoms of xanthogranulomatous cholecystitis are unspecific, similar to common cholecystitis. Furthermore, there are no significant differences in signs and symptoms between xanthogranulomatous cholecystitis and gallbladder cancer.

In the present case, the authors demonstrate the diagnostic dilemma due to intraoperative misdiagnosis by both macroscopically gallbladder findings and frozen-intrassection analysis. One patient that a clinical history of biliary colic and abdominal ultrasound showing multiple gallstones was presented, which mimicked gallbladder cancer at intraoperative evaluation. The main symptom at presentation suggested cholecystolithiasis and patient was admitted for laparoscopic cholecystectomy.

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Imaging techniques such as ultrasonography, computed tomography or even magnetic resonance imaging can present limitation for differentiating either benign or malign lesions into gallbladder. Computed tomography findings such as continuous mucosal line in a diffuse gallbladder wall thickening, intramural hypoattenuated nodule (xanthogranulomas composed by foamy histiocytes, lymphocytes and multinucleated giant cells), the presence of gallstones and pericholecystic infiltration have been significantly associated with xanthogranulomatous
cholecystitis. Moreover, focal gallbladder wall thickening was observed more frequently in gallbladder cancer. 11-13

In the present case, intraoperative findings revealed gallbladder wall thickening and adhesion of the gallbladder to the liver. Current literature showed the major intraoperative findings of xanthogranulomatous cholecystitis included thickening of the gallbladder wall (90%), and adhesions of the gallbladder to adjacent tissues and organs (87%). 1 The impossibility to exclude gallbladder cancer lead us to conduct a frozen-section investigation, that diagnosed a malignant lesion, indicating a more aggressive surgical approach. Locally advanced gallbladder carcinoma can be frequently confounded with xanthogranulomatous cholecystitis. However, only radical resection with free margins which generally involves major hepatectomy may offer the cure of the locally advanced gallbladder carcinoma. 16 In Brazil, Pais-Costa et al 15 showed that major hepatectomy plus hilar lymphadenectomy for treating locally advanced gallbladder carcinoma may lead to long-term survival in very selected cases. Therefore, the present patient underwent a inferior central hepatic bisegmentectomy (hepatic resection of the segments IV and V) plus hilar lymphadenectomy as described by Costa et al, 16 the aim of this surgery was to obtain long-term survival in a case which intraoperative frozen section revealed resectable gallbladder carcinoma. On the contrary that observed by Pais-Costa et al, 15 postoperative complications such as infection of incisional wound, hepatic failure, pleural effusion, seroperitoneum, biliary fistulas and cholangitic stenosis were not observed in this case.

According to the literature, 19% is the conversion rate to open cholecystectomy in patients with xanthogranulomatous cholecystitis. 8 Laparoscopic procedure is difficult to be performed in xanthogranulomatous cholecystitis patients because of the high frequency of complications such as gallbladder perforation, biliary fistulas, hepatic abscesses and extended inflammation into adjacent structures. 8,9 If xanthogranulomatous cholecystitis is suspect, a frozen-section examination should be done because of the difficulty to differentiate macroscopically xanthogranulomatous cholecystitis from gallbladder carcinoma or even considering the possibility of coexistence of both disorders. Even when performing a frozen-section examination, the correct diagnosis can still be missed maybe due to the presence of inflammatory atypia in the epithelium. 10

In conclusion, xanthogranulomatous cholecystitis is often misdiagnosed pre-operatively and intra-operatively as having gallbladder cancer, leading to an unsuccessfully laparoscopic procedure, a high conversion rate and a more aggressive surgical resection. The authors like observed in previous reports, warrant further investigations on the diagnosis of xanthogranulomatous cholecystitis to avoid unnecessary extensive surgery to treat this benign inflammatory condition.

DISCLOSURE
No potential conflict of interests was reported

REFERENCES


