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Xanthogranulomatous Cholecystitis: A Rare form of Chronic Cholecystitis

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Abstract **Case Report**

Xanthogranulomatous cholecystitis (XGC) is a rare form of cholecystitis distinguished by the presence of numerous yellow-brown intramural nodules with severe fibrosis and foam cells within the gallbladder wall, which may be misdiagnosed as a Gall Bladder Carcinoma. It is typically diagnosed between the sixth and seventh decades of life, predominantly in females. Clinical signs and symptoms are comparable to those of acute and chronic cholecystitis, making it challenging to distinguish XGC prior to surgery. The case of a 52-year-old man who presented to the emergency department with right upper quadrant abdominal pain for 4 months, radiating into the right shoulder is discussed. Patient was diagnosed to have Chronic Calcular Cholecystitis on Ultrasound. Laparoscopic Cholecystectomy was performed and XGC was diagnosed post-operatively through definitive histopathological examination. Preoperatively and intraoperatively, Xanthogranulomatous Cholecystitis is difficult to diagnose and pathological examination is required for a definitive diagnosis. XGC, which exhibits clinical and radiological signs similar to those of gallbladder tumors, must be included in the differential diagnosis in order to avoid an extensive surgical procedure.

Keywords: Gall Bladder, Xanthogranulomatous Cholecystitis, Gall Bladder Cancer, Cholecystitis, Laparoscopic Cholecystectomy, Gall Stones, Cholelithiasis, Chronic Cholecystitis.

INTRODUCTION

Xanthogranulomatous cholecystitis (XGC) is a rare type of cholecystitis that manifests with non-specific symptoms and characteristics, making it difficult to differentiate between chronic cholecystitis and Gall Bladder Cancer. Typically, XGC is a histological diagnosis for focal or diffuse acute and chronic cholecystitis.

McCoy *et al.*, coined the terminology in 1976, though Christensen *et al.*, initially described it in 1970 [2, 3]. Christensen et al. and Amazon *et al.*, described a pseudotumoral subtype of chronic cholecystitis distinguished by xanthoma-like foam cells, scarring, with ceroid (wax-like) nodules in the inflamed gallbladder wall. They used the terminology fibroxanthogranulomatous inflammation and gallbladder ceroid granulomas, which today are synonymous with XGC [3, 4].

Preoperative imaging is beneficial, but the diagnosis is frequently a postoperative histopathological discovery. For these patients' surgical management to be effective, a precise diagnosis is critical. Making a distinction between XGC and Gall Bladder Cancer (GBC) preoperatively would assist to prevent unnecessary morbidity, particularly in the form of extensive surgery.

CASE REPORT

A 52-year-old male came to the emergency unit with a one-week history of severe abdominal pain. The pain originated in the right upper quadrant and radiating to the epigastrium and right shoulder. The patient began to experience pain four months ago, and its severity had increased during the past two months. The discomfort was severe, increased by meal consumption, and alleviated by antacid medications. Pain was accompanied by nausea and numerous episodes of vomiting. There were no bouts of fever, and his bowel and bladder habits remained unchanged. No history of anorexia or weight loss was reported. Patient had multiple visits to the local general practitioner and was prescribed analgesics and proton-pump Inhibitors.

The patient's medical history includes hypertension and diabetes mellitus. There were no prior surgical procedures. There was no smoking or alcohol consumption history.

Ultrasound Abdomen (Figure 1) reported Increased Wall Thickness of Gall Bladder with a 1 cm stone at the neck, indicating a pre-operative diagnosis of Chronic Calcular Cholecystitis. The decision was made to perform Laparoscopic Cholecystectomy. Total Cholecystectomy was successfully performed without any intraoperative complications. The patient began oral feeding on the first postoperative day and was discharged to home on 2nd post- operative day. Histopathological analysis of the resected specimen revealed lipid-laden macrophages and chronic inflammatory characteristics. These results led to the diagnosis of Xanthogranulomatous cholecystitis (Figure 2). Two weeks later, the patient was seen in OPD and indicated full recovery with no post-operative issues.

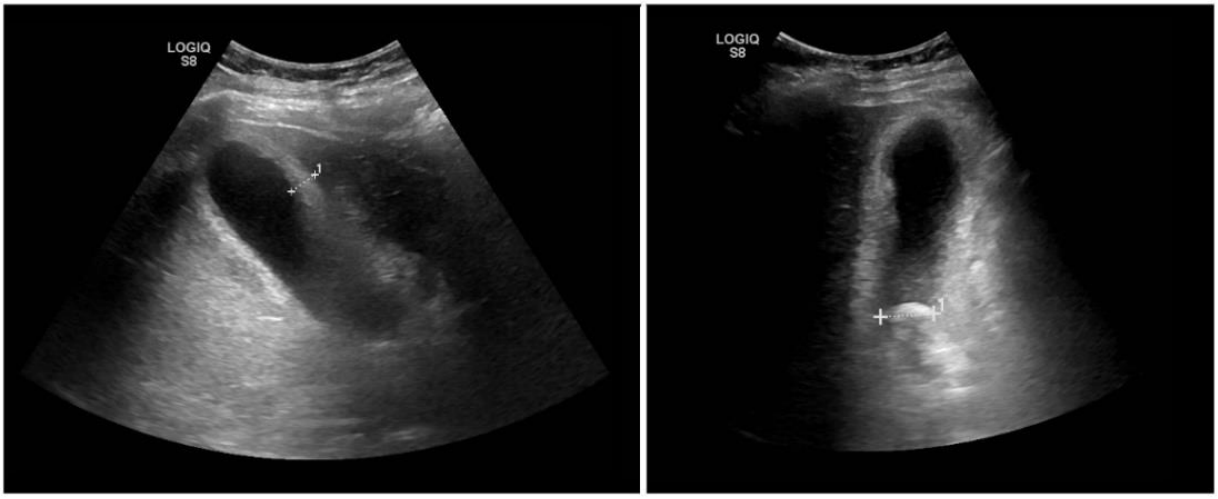

Figure 1: Increased Wall Thickness of Gall Bladder (L), Stone in the Neck of the Gall Bladder (R)

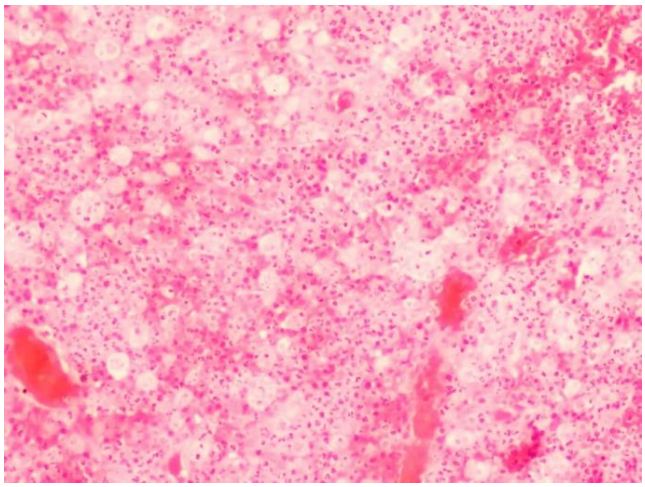

Figure 2: Histopathological analysis reveals lipid-laden macrophages alongside chronic inflammatory cells

DISCUSSION

XGC is a rare gallbladder disease characterized by significant fibrosis and a thickening of the gallbladder's wall. XGC is defined histologically by variable degrees of chronic or acute inflammatory cell infiltration, the presence of numerous macrophages containing lipids, and, in the advanced stages, fibrosis. Macroscopically, the lesions range from a small, localized center within a yellow-brown nodule in the gallbladder wall to diffuse involvement of the complete gallbladder with extension into adjoining tissues [1, 2].

In 1981, Goodman and Ishak coined the term Xanthogranulomatous cholecystitis after reviewing 40 cases from the Armed Forces Institute of Pathology [3]. A small percentage of cholecystectomy specimens include XGC, ranging from 0.7% to 1.8% in the United States [4, 5], 9.3% to 13.2% in India [6, 7], and 1.2% to 10% in Japan [9, 10]. Although the pathophysiology of XGC is unknown, gallstone blockage and stasis are key etiologic factors [10].

Current evidence suggests that the lesion begins with inflammation and blockage of the gallbladder wall. The bile then enters the gallbladder

stroma through ulcerations in the surface mucosa or ruptured Aschoff- Rokitansky sinuses. At the site of inflammation, macrophages consume bile lipids to create big, spherical, pale xanthoma cells [5, 9]. The extravasation of bile into the gallbladder wall causes lipid-containing histiocytes to infiltrate the outer layer of the muscle lining the gallbladder wall, producing Xanthogranulomatous foci and fibrosis [3, 5]. XGC can be discovered inadvertently during elective cholecystectomy and is frequently misdiagnosed as gallbladder malignancy.

The coexistence of XGC and gallbladder cancer is an added complication [3, 10]. Nearly 10% of cases coexist in this way. In Japan and the United States [3, 9], 0.2% in India, and the majority of reported cases were identified by histologic study of cholecystectomy specimens [12, 13]. Since serum carbohydrate antigen (CA19-9) levels are elevated in both gallbladder cancer and XGC, the tumor marker CA19-9 is not helpful for the diagnosis of coexisting carcinoma or for differentiating XGC from carcinoma [14-16].

Ultrasonography assists in the identification of focal or diffuse gallbladder wall thickening, cholecystolithiasis, and intramural hypoechoic nodules. Lee observed that some ultrasonographic characteristics suggested XGC, including gallbladder wall thickening, cholecystolithiasis, and infiltration into surrounding tissues [17]. An abdominal CT can also confirm these abnormalities. On CT, typical manifestations of XGC include diffuse gallbladder wall thickening, intramural hypodense nodules, pericholecystic infiltration, and hepatic abscess [18]. In one study, nearly 33% of patients had intramural hypodense nodules, which was one of the most specific CT findings for XGC [19].

Also, observable is the involvement of the biliary tract in the inflammatory process. On the other hand, the absence of intrahepatic biliary dilatation is more prevalent in XGC and is a crucial characteristic for distinguishing gallbladder carcinomas [20]. Although current research suggests that MRI is more accurate than CT and high-resolution ultrasound in identifying XGC, the presence of (diffuse wall thickening, hypodense bands or intramural hypoattenuating nodules, gallstones, and ceaseless mucosal line) are independently associated with XGC on high-resolution ultrasound and CT. However, the hallmarks of XGC, such as adhesions, thickening of the gallbladder wall, and pericholecystic fat infiltration, might substantially resemble those of adenomyosis and Gall bladder cancer [21]. Microscopic examination indicates variable amounts of inflammation-forming foamy histocytes, big cells, and fibrosis, but no dysplastic or malignant change [17].

XGC also is associated with an increased rate of conversion from laparoscopic to open operative procedures owing to procedural difficulty related to local inflammation creating dense adhesions or gallbladder wall thickening [23- 28].

Determining the definitive diagnosis in such these cases can be a difficult task, and it is often deferred until the final pathology reports are available. Due to the fact that both XGC and GBC share comparable clinical, imaging, and intraoperative characteristics, it is difficult to determine the correct diagnosis and approach.

CONCLUSION

Xanthogranulomatous cholecystitis, despite its characteristic histopathological presentation, the clinical and radiological presentation, is nonspecific and difficult to differentiate from other forms of cholecystitis and sometimes from gallbladder cancer. Therefore, it is difficult to diagnose XGC prior to surgery. Prevalent adhesions of the gallbladder to adjacent organs and tissues make surgical procedures challenging. Since frozen-section analysis plays an important role in determining the nature of the lesions, intraoperative frozen-section examination should be performed to differentiate XGC from gallbladder carcinoma, and when a suspicious finding is observed, the pathologist may direct the surgeon to perform a necessary resection.

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