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Xanthogranulomatous Cholecystitis - An Uncommon Variant

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Authors' contributions

This work was carried out in collaboration among all authors Author GSJ designed and produced majority of the manuscript. Author RSP was the supervising surgeon for this patient. Author GA obtained all clinical images for and was involved in the drafting of manuscript. All authors were directly involved in managing this patient.

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Case Report

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ABSTRACT

Aim: Xanthogranulomatous cholecystitis is a rare variant of inflammatory disease of gallbladder which is marked by the presence of lipid-laden macrophages along with extensive fibrosis. It is frequently confused with carcinoma of the gallbladder preoperatively on imaging or on gross appearance intraoperatively. Definitive diagnosis is confirmed on histopathological examination. Imaging may shed light on this condition preoperatively but may a time this may present as a histological surprise. Accurate preoperative diagnosis is necessary to avoid radical resection and morbidity in patients.

We discuss here a case of 36-year-old female who presented with abdominal pain. Her CT scan of abdomen showed borderline thickening of gallbladder wall and chronic cholecystitis however on histopathological report was suggestive of xanthogranulomatous cholecystitis.

Keywords: Xanthogranulomatous cholecystitis; carcinoma GB; gallbladder; cholecystectomy.

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1. INTRODUCTION

"Xanthogranulomatous cholecystitis (XGC) is a rare variety of chronic inflammatory diseases of the gallbladder (GB). It is characterized by a focal or diffuse inflammatory process with marked proliferative fibrosis and infiltration of lipid-laden macrophages. The incidence of XGC ranges from 0.7 to 10%" [1,2]. It is often misdiagnosed as malignancy of gall bladder (GB) on imaging and intraoperative findings.

"The most important association of XGC is with gallstones which are seen in as many as 80% of cases. It presents with non-specific symptoms and signs, making it indistinguishable from typical acute or chronic cholecystitis. XGC is a histopathological diagnosis of focal or diffuse acute and chronic cholecystitis. Microscopically, it is hallmarked by the presence of lipid-containing histiocytes infiltrating into the outer layer of the muscle lining the gallbladder wall may be seen to form xanthogranulomatous foci and fibrosis owing to extravasation of bile into the gallbladder wall" [1,3,4].

We present a case report of a patient who presented with features of chronic cholecystitis.

2. PRESENTATION OF CASE

A 36-year-old female with no prior medical history presented to our outpatient department with progressive pain in the right hypochondrium for 4 months. The pain was moderate in intensity, intermittent, and radiating to the back with no association to food intake, aggravating or relieving factors. No history of fever, chills, nausea, vomiting or weight loss was reported. There were no associated bowel or bladder complaints. Past surgical and family history was unremarkable. At the initial examination, the vital parameters were within normal limits. The abdomen was soft with no tenderness, guarding, or rigidity. No mass was palpable on the physical examination. The rest of the systemic examination was within normal limits.

All routine blood investigations were within the normal limit. Abdominal ultrasound showed diffusely thickened gallbladder wall with features of chronic cholecystitis. Computed Tomography (CT-scan) further confirmed the presence of chronic acalculous cholecystitis with no dilatation of the common bile duct (Fig. 1). Laparoscopic cholecystectomy was carried out and the specimen was sent for histopathological examination. Gross examination (Fig. 2) demonstrated a 6.8 cm long specimen, with the greatest diameter of 3.1 cm. The gall bladder was increased in thickness and the external surface appeared congested. The mucosa was flattened and showed ulceration. The lumen contains bile and necrotic material. Histological examination revealed xanthogranulomatous cholecystitis (Fig. 3), section showed mucosal lining and underlying aggregates of histiocytes in submucosa and muscularis mucosa with no evidence of malignancy. The patient had an uneventful post-operative hospital stay and was discharged in stable condition on the 4th postoperative day. The patient was followed up as per standard norms. She recovered well and is in good health.

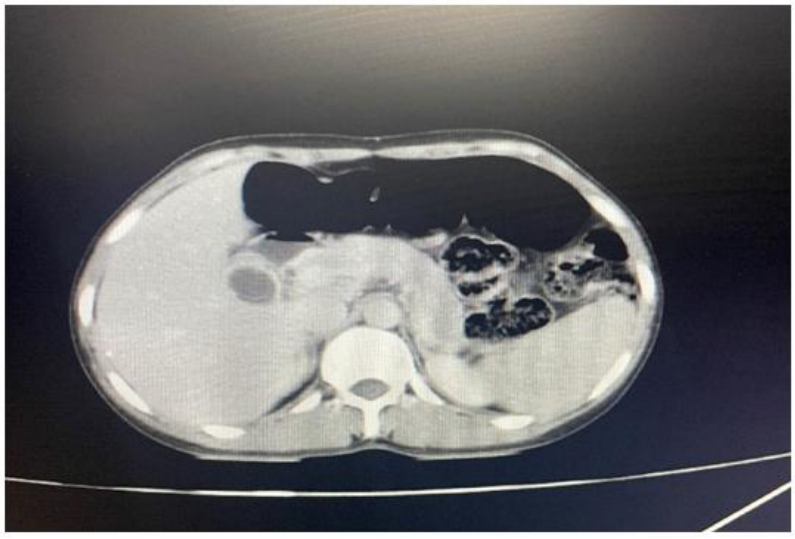

Fig. 1. CT scan axial view showing thickened gall bladder

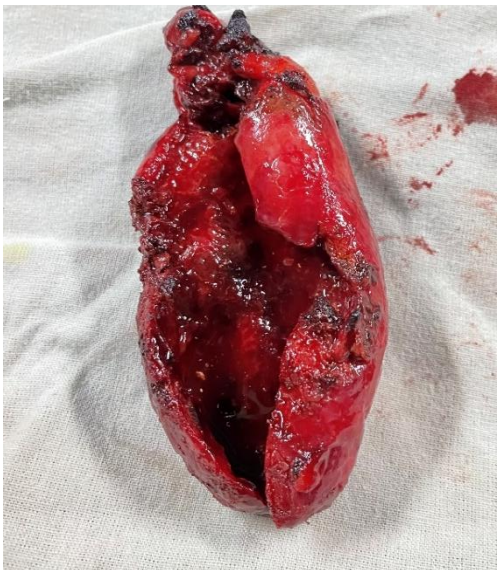

Fig. 2. Gross appearance of the cut open specimen of gallbladder

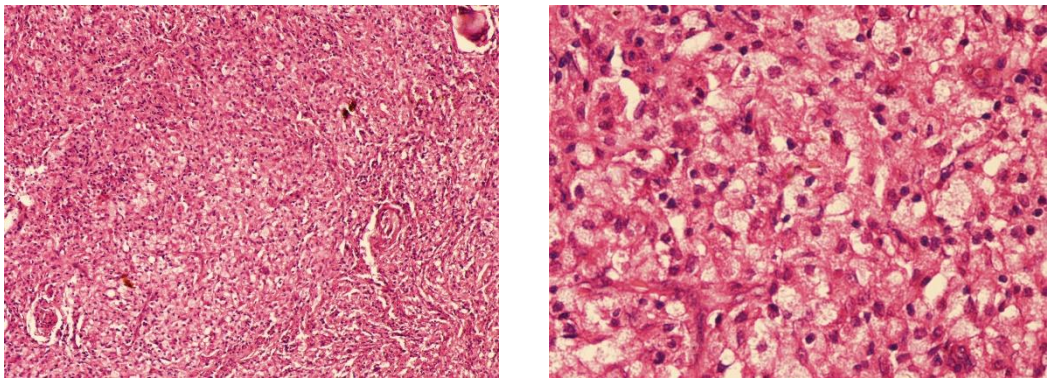

Fig. 3. Histopathology slide showing cholecystitis next showing a varying degree of inflammation with presence of giant cells, foamy histiocyte cell and fibrosis without any dysplasia

3. DISCUSSION

"Xanthogranulomatous cholecystitis is a rare form of chronic cholecystitis with incidence ranging from 1.3% to 5.2% of resected GB specimens. It is more common in male patients with a male to female ratio of 2:1" [1,4]. "Gallstones are one of the most important associations of XGC which is seen in up to 80% of cases" [3]. "A previous study concluded that there may be a significant geographical influence on the incidence of XGC with rates as many as three to four times higher in India. This may be accounted for a high incidence of gallstones in India" [5]. "XGC is very often misdiagnosed as gallbladder carcinoma (GB Ca) on clinical presentation, ultrasound, computed tomography (CT), and intra-operative findings. The incidence of a co-existing carcinoma in patients of XGC varies from 2-5 %" [6-9].

"The mechanism which results in this condition remains unclear, but it has been postulated that XGC begins as a biliary obstruction with acute or chronic cholelithiasis which results in increasing intra-gallbladder pressure. It is thought that this rise in pressure provokes a rupture of the Rokitansky-Aschoff sinuses or results in the formation of mucosal ulcer with extravasation of bile in the interstitial tissues and therefore resulting in a xanthogranulomatous inflammatory reaction" [4,6].

"The histopathological diagnosis is based on the appearance of diffuse or focal mural changes in the form of xanthoma cells (which are foamy

histiocytes containing lipids and bile pigment), giant multinucleate histiocytes, and acute or chronic inflammatory cells. These pathognomonic histiocytes are positive for CD68 in immunohistochemistry studies. There is an associated formation of microabscesses in the gallbladder wall and a consequent fibrous reaction and scarring. Rupture of gallbladder serosal lining results in the spread of the inflammatory response consequently leading to adhesions with adjacent liver, duodenum, and transverse colon" [10].

"The clinical spectrum of XGC usually involves acute or chronic cholecystitis including right hypochondriac pain which may be radiating to the shoulder and back, fever, nausea, and vomiting; thereby making XGC indistinguishable from cholecystitis clinically" [11]. "It is to be noted that abdominal pain, jaundice, and fever are more frequently seen as a presenting feature in patients with XGC as compared to patients with GB cancer" [12]. "The sonographic findings seen in patients with this ailment include the presence of gallstones or sludge and moderate to marked focal or diffuse thickening of the gallbladder wall. Parra et al" [11] "observed that the wall thickening was hyperechoic in comparison to the liver in 100% of patients. Xanthogranulomatous nodules are seen as well-defined hypoechoic areas on ultrasound. Hypoechoic bands might be caused by more generalized involvement of the mucosa of the gall bladder. Complications like perforation, abscess, and hepatic infiltration are identified on ultrasound. Computed tomography (CT) findings of patients presenting with acute symptoms are similar to those presenting with chronic symptoms. CT findings include – a wide spectrum of lesions which include diffuse or focal mural thickening, intramural hypoattenuating nodules seen in thickened gall bladder walls (halo sign), luminal surface enhancement (LSE) with continuous mucosal lines or mucosal lines with a focal breach, intra-hepatic duct dilatation and a loss of interface between the gallbladder and the liver. Cholelithiasis and choledocholithiasis are seen associated with XGC" [10,11].

"A pre-operative ultrasound and CT in patients with XGC may display above mentioned specific features which are suggestive of XGC. These features are also seen in GB Ca, but visualization of a continuous mucosal line and the absence of pericholecystic fluid, both suggest a diagnosis favoring XGC rather than GB Ca. This may be helpful to differentiate between XGC and GB Ca" [11]. "XGC is more likely mistaken for GB cancer macroscopically than radiologically, especially in patients with XGC with severe proliferative fibrosis involving the gall bladder and the surrounding organs. A gross examination of the mucosa along with the frozen section can aid in differentiating XGC from GB cancer and exclude their simultaneous presence" [12].

"It is now estimated that XGC and GB cancer coexist in up to 12% of cases. Therefore, even if a preoperative diagnosis is made with fine-needle aspiration cytology, it is crucial to be aware of the fact that there may be a possible coexistence of XGC and cancer in the same GB. Zhuang et al demonstrated the relation of BCL-2 and c-Myc oncogenes with XGC, thereby establishing its precancerous nature. Hence, in addition to several frozen section examinations, a vigilant gross observation during surgery is needed even if the pre-operative diagnosis of XGC has been made" [12].

"An increased rate of conversion from laparoscopic to open operative procedures have been found in cases of XGC. This can be attributed to procedural difficulty secondary to local inflammation-causing dense adhesions with gallbladder wall thickening" [1,11, 13,14].

4. CONCLUSION

The pseudo tumoral nature of XGC has puzzled surgeons in terms of surgical treatment. Even with the use of modern imaging techniques, a clear differential diagnosis between XGC and malignant GB lesions is often challenging. The intraoperative differential diagnosis of XGC from GB carcinoma is difficult when XGC is associated with tumor formation and adhesions to adjacent organs, and a definitive diagnosis necessitates a histopathological examination. An accurate preoperative diagnosis based on clinical and radiological features may help in avoiding radical resection and morbidity in patients.

CONSENT

All the authors declare that 'written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images'.

ETHICAL APPROVAL

As per international standard guidelines, written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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