Radiology Case Reports 19 (2024) 5674–5677

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Navigating diagnostic challenges in xanthogranulomatous cholecystitis: A case report ☆

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ARTICLE INFO

Article history:
Received 5 August 2024
Revised 9 August 2024
Accepted 10 August 2024

Keywords:
Adenomyomatosis
Cholecystectomy
Gall bladder
Xanthogranulomatous cholecystitis

ABSTRACT

Xanthogranulomatous cholecystitis (XGC) presents a diagnostic challenge due to its rarity and varied clinical manifestations and nonspecific radiological findings. We here describe a 67-year-old man with right hypochondriac pain, where imaging revealed irregular thickening of the gallbladder wall, prompting consideration of various differential diagnoses including gallbladder malignancy, adenomyomatosis, and complicated cholecystitis. With inconclusive lab results, cholecystectomy with potential extended hepatectomy was advised. Intraoperatively, an inflamed gallbladder was observed. Histopathological examination confirmed XGC, stressing histological verification. Complete cholecystectomy is standard, with partial resection an option. Our case details the complexity in diagnosis and management of XGC.

Introduction

Xanthogranulomatous cholecystitis (XGC) is a rare, benign, chronic inflammatory condition affecting the gallbladder, categorized as an uncommon variant of chronic cholecystitis [1]. The prevalence of XGC varies depending on the geographic region, with rates ranging from 1.3% to 1.9% in Western nations such as Europe and the USA, while in India, it has been reported to be as high as 9% [2].

XGC usually presents similarly to acute cholecystitis, featuring symptoms like nausea, vomiting, right upper quadrant pain, and a positive Murphy's sign, but it can also exhibit a more chronic course with weight loss and anorexia. Ultrasound (US) may reveal intramural hypoechoic nodules or bands and diffuse gallbladder wall thickening, while

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.08.050

computed tomography (CT) and magnetic resonance imaging (MRI) show similar findings along with pericholecystic infiltration and hepatic attenuation differences. Complications, seen in about 30% of cases, include gallbladder perforation, bile duct obstruction, liver abscesses, and fistulas. Treatment typically involves open cholecystectomy, although laparoscopic attempts are made initially, despite the higher complication rate associated with this approach. Differentiating XGC from other conditions like gallbladder carcinoma is challenging due to its nonspecific clinical presentation and radiological features [3].

Case report

We present the case of a 67-year-old man who sought medical attention at the surgery outpatient clinic due to right hypochondriac abdominal pain, prompting suspicion of liver and gall bladder issues. An abdominal US was performed, revealing localized irregular thickening of the gall bladder wall in the body region, approximately measuring 2.5 cm, along with luminal narrowing (Fig. 1). Additionally, linear hyperechogenicities and adjacent hypoechogenicity were observed in the liver parenchyma contiguous with the thickened gall bladder wall (Fig. 2). The differential diagnoses considered included gall bladder malignancy, adenomyomatosis, and complicated cholecystitis. Laboratory findings were nonconclusive.

Further evaluation through contrast-enhanced computed tomography (CECT) of the abdomen and pelvis was conducted, confirming focal thickening of the gall bladder wall anteriorly, with areas of fatty attenuation not showing enhancement on postcontrast phases of the study (Fig. 3). The adjacent liver parenchyma exhibited heterogeneity on both noncontrast and postcontrast studies (Fig. 4). Mild pericholecystic fat stranding and an enlarged peripancreatic lymph node were also observed (Fig. 5).

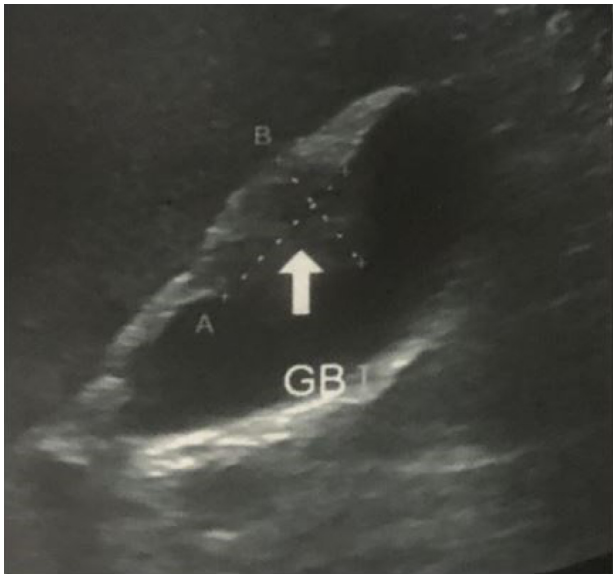

Fig. 1 – Ultrasound study shows localized irregular thickening of the gall bladder wall (vertically oriented arrow) in the body region with luminal narrowing.

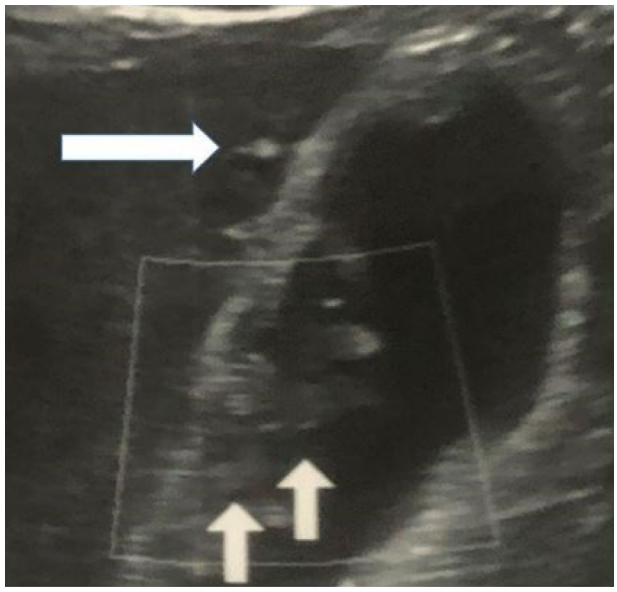

Fig. 2 – Ultrasound study shows linear hyperechogenicities with adjacent hypoechoic areas in the liver parenchyma (horizontally oriented arrow) contiguous with the thickened gall bladder wall (vertically oriented arrow).

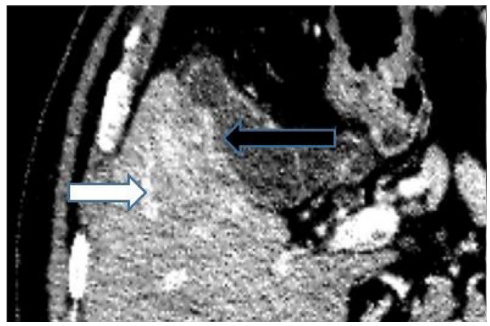

Fig. 3 – CECT study shows thickening and enhancement of the posterior wall of gall bladder (white arrow). Heterogeneous post contrast enhancement noted in the liver parenchyma adjacent to the gall bladder wall thickening (black arrow).

Given the possibility of gall bladder malignancy and adenomyomatosis, the recommendation for cholecystectomy with potential extended hepatectomy was made. During the surgical procedure, inflamed gall bladder and pericholecystic adhesions were encountered and complete cholecystectomy was performed with possible diagnosis of empyema gall bladder.

Subsequent histopathological examination revealed a thickened gall bladder with infiltration of various immune cells and sheets of foamy cells containing brownish pigments, indicative of XGC of the gall bladder wall (Figs. 6-8).

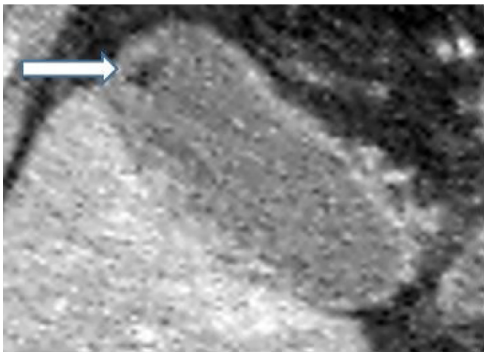

Fig. 4 – CECT study shows fundal wall thickening with areas of fat attenuation (white arrow) and nonenhancement on post contrast study.

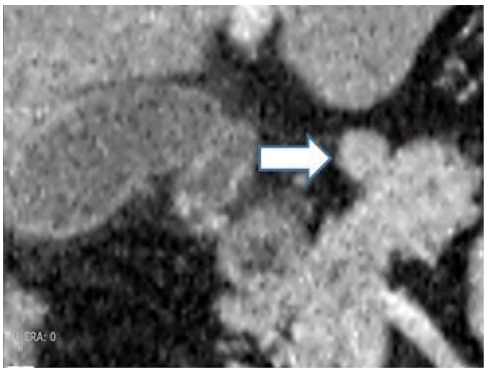

Fig. 5 – CECT study shows a homogeneously enhancing lymph node in the peripancreatic region.

Discussion

XGC is a rare gallbladder inflammation characterized by the accumulation of lipid-laden macrophages in response to leaked bile. Its incidence is low, affecting approximately 1.46% of cholecystectomy cases, with a slight male predominance. The exact cause of bile leakage leading to XGC is unclear, but it may result from gallstone-related mucosal damage or ruptured Rokitansky–Aschoff sinuses. Clinical symptoms include right upper quadrant pain, nausea, vomiting, and weight loss. Diagnosis is challenging as serological evaluation and tumor markers like CA 19-9 are often nonspecific and physical examination may reveal a positive Murphy's sign and a right upper quadrant mass [4].

US is valuable for detecting gallbladder wall thickening, cholelithiasis, and intramural hypoechoic nodules, which can be indicative of XGC. These features, including thickened gallbladder wall and infiltration into adjacent tissues can be observed on ultrasonography. Abdominal CT can further confirm these findings, showing diffuse gallbladder wall thickening, intramural hypodense nodules, pericholecystic infiltration, and hepatic abscess. Notably, intramural hypodense nodules are particularly specific to XGC on CT, with around 33% of patients exhibiting this characteristic [5].

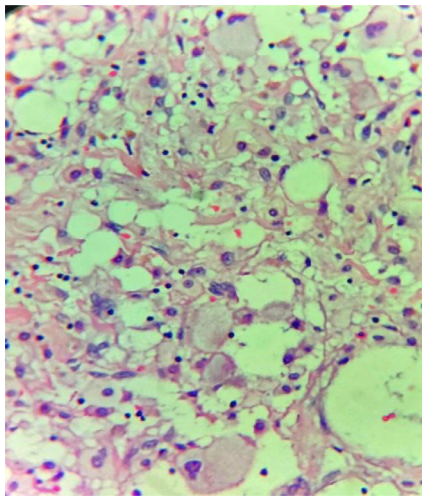

Fig. 6 – Showing sheets of foamy cells.

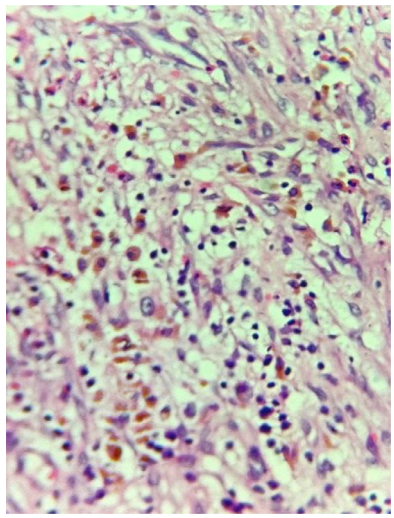

Fig. 7 – Brown colored pigments in the gall bladder wall.

Distinguishing XGC from other inflammatory or malignant conditions before surgery is challenging due to other aggressive pathologies, necessitating histological confirmation. Macroscopic examination typically reveals gallbladder wall thickening, fibrous adhesions, fistulous tracts, and ceroid yellowed nodules [4]. The microscopic findings used to identify XGC in the pathology samples include the presence of histiocytes, cholesterol accumulations, lipids, and either focal or diffuse wall enlargement. Additionally, foreign body giant cells or multinucleated giant cells, cells engulfing lipids and bile pigments leading to the formation of xanthomatous cells, as well as the existence of acute and chronic inflammatory cells, are also evident [6]. As macroscopic features overlap with malignant processes it complicates the diagnosis thus histopathological evaluation becomes crucial.

Intraoperative frozen section biopsies serve as a valuable resource for differentiating diagnoses and assisting specialists in making informed decisions. However, managing XGC

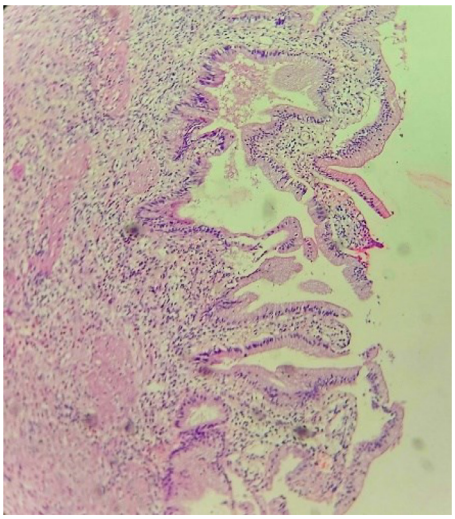

Fig. 8 – Gall bladder wall infiltration with lymphocytes, polymorphs, eosinophils and neutrophils.

is still complex, and diagnostic uncertainties can cause substantial challenges for patients and their families, potentially resulting in dilemma in the course of treatment. Nonetheless, simple cholecystectomy is typically the preferred treatment for XGC [7]. Although complete cholecystectomy is the preferred approach for treating XGC due to its potential association with gall bladder carcinoma, partial cholecystectomy might be considered when complete resection is not feasible. In some cases, hepatectomy or pancreaticoduodenectomy may be necessary if there is involvement of the nearby liver or pancreatic tissues [5].

Conclusion

In summary, this case underscores the diagnostic challenges associated with XGC. Presenting with symptoms of vague abdominal pain in right hypochondriac region, the imaging findings revealed atypical thickening of the gallbladder wall, prompting consideration of various potential diagnoses, including cancer. Surgical intervention, in the form of cholecystectomy, was pursued due to suspected malignancy, with subsequent histopathological examination confirming the presence of XGC. This emphasizes the importance of a collaborative approach in accurately diagnosing and managing XGC.

Patient consent

Written informed consent for publication of their clinical details and/or clinical images were obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

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