



A CASE OF XANTHOGNULOMATOUS CHOLECYSTITIS WITH SIGNIFICANTLY ELEVATED LEVELS OF SERUM CARBOHYDRATE ANTIGEN 19-9 - A RARE CASE REPORT

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ABSTRACT Serum carbohydrate antigen 19-9 (CA 19-9), a marker of malignant tumors, is generally slightly elevated in benign conditions. We report a case of xanthogranulomatous cholecystitis with a significantly elevated level of serum CA 19-9 levels. A 27-year-old woman presented with abdominal pain, intermittent vomiting, jaundice and loss of appetite. Ultrasonography, contrast-enhanced computed tomography and magnetic resonance cholangiopancreatography were carried out which revealed an irregular gallbladder wall thickening with loss of gall bladder hepatic interface. The serum CA 19-9 level was significantly elevated to 7590 U/ml. The patient then underwent exploratory laparotomy and cholecystectomy 12 days after admission. Intraoperative frozen section was performed to rule out malignancy and diagnosis of xanthogranulomatous cholecystitis was made. Subsequently histopathological examination was done and the diagnosis of xanthogranulomatous cholecystitis was confirmed. The levels of Carbohydrate antigen 19-9 returned to normal post surgery. Xanthogranulomatous cholecystitis with a significantly high elevation of the serum CA 19-9 level is rare. In such cases, it is important to confirm and distinguish between inflammation and malignancy.

KEYWORDS : Cholecystitis, Carbohydrate antigen 19-9, Xanthogranulomatous Cholecystitis

INTRODUCTION:

Serum carbohydrate antigen 19-9 (CA 19-9), is a marker of malignant tumors. The levels are often high in biliary tract cancers and pancreatic cancers. In some cases, CA19-9 levels can also be high in benign conditions like cholangitis and pancreatitis.¹

Moderate elevations of CA 19-9 may be seen in benign disease, but levels in the thousands are indicative of malignancy.² We report a case of xanthogranulomatous cholecystitis in a young patient with a significantly elevated level of serum CA 19-9 levels. The levels of serum CA 19-9 returned to normal post surgery.

Case Report:

A 27 yrs old patient came to the department of surgical oncology complaining of pain abdomen, intermittent vomiting, jaundice and loss of appetite since 5 months.

On per abdominal examination, a tense cystic mass was noted in the right hypochondrium of size 4 x 4 cm. Ultrasonography whole abdomen was done which revealed cholelithiasis, choledocholithiasis with upstream biliopathy. Alkaline phosphatase level was raised, 759 U/L. Serum CEA level was normal. Serum CA 19.9 was found to be markedly increased, 7590 U/ml.

MRCP was performed which revealed calculus in GB neck, multiple cystic spaces in GB wall, enhancing wall thickening with loss of GB hepatic interface. CECT abdomen reveals irregular asymmetrical wall thickening involving the fundus and body of gall bladder with loss of GB hepatic interface.

The patient then underwent exploratory laparotomy and cholecystectomy. Intraoperative frozen section was performed to rule out malignancy where diagnosis of xanthogranulomatous cholecystitis was made.

Subsequently Histopathological examination was done and diagnosis of xanthogranulomatous cholecystitis was confirmed. The levels of

Carbohydrate antigen 19-9 returned to normal immediately after surgery.

Radiological Imaging:

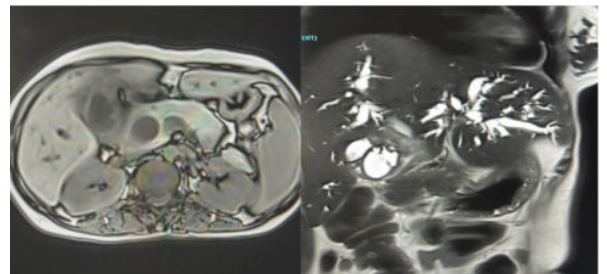


Fig (a), (b): Imaging study showing gall bladder wall thickening

Gross Findings:

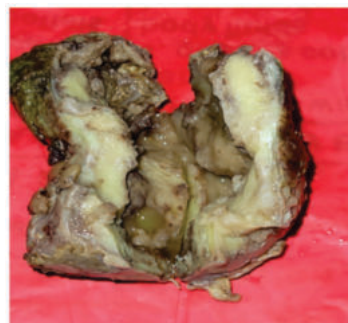


Fig (c): Photomicrograph showing irregularly thickened gall bladder wall grossly

Frozen findings:

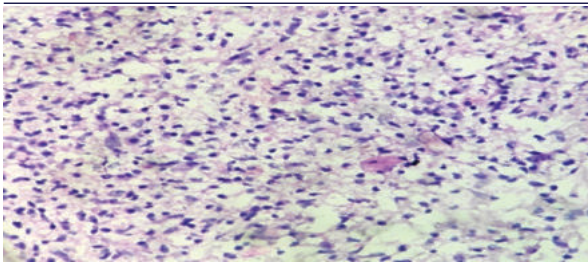


Fig (d): Photomicrograph showing xanthoma cells in frozen section

Histopathological Examination:

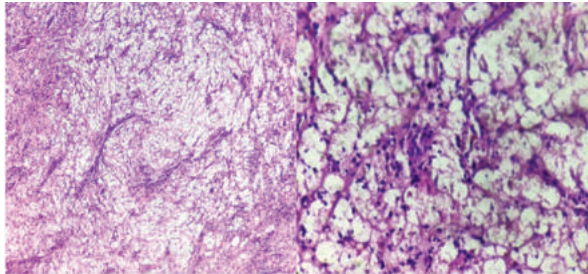


Fig (e): Lower power view showing lipid laden macrophages and inflammatory cells

Fig (f) High power view showing lipid laden macrophages and infiltrate of inflammatory cells

DISCUSSION:

CA19-9 is a carbohydrate antigen which is found to be elevated in patients with cholangiocarcinomas, pancreatic cancers and also gastrointestinal cancers. It is used for the diagnosis as well as for recurrence of a malignant tumor. The levels of CA19-9 may also be elevated in benign diseases like cholangitis and pancreatitis; but levels higher than 1,000 U/ml can be rarely seen.¹

Xanthogranulomatous cholecystitis is a rare form of cholecystitis, characterized by the presence of xanthogranuloma which are prominent yellow structures within the gallbladder wall that is very often lithiasic. It can mimic gallbladder carcinoma when it presents in its pseudo-tumoral form with occasional adjacent organ involvement. Its association with gall bladder carcinoma remains a topic of debate in the literature. Biopsy may be necessary to eliminate the diagnosis of tumor. CA 19.9 can be elevated several thousand fold during xanthogranulomatous cholecystitis. It is often measured in case of diagnostic doubt, but is not conclusive of malignancy.³

Ruling out malignancy is important in case of elevated CA19-9 in the presence of cholelithiasis or cholecystitis to determine the course of treatment. In cases where diagnosis is difficult, other modalities such as endoscopic ultrasonography may be considered for evaluation, including drainage and cytological diagnosis.⁴

The abnormally high levels of CA19-9 found in our case was interesting. Elevated levels of CA19-9 can also be found in benign diseases such as cholangitis. Also ultrasonography showed a thickening of the bile duct wall, the possibility of the influence of the mild cholangitis exhibited by the patient could not be ruled out. But elevated CA19-9 levels reaching above 1,000 U/ml are rare in benign diseases. Only few cases were found in literature where abnormally high levels of CA19.9 was seen in benign conditions.

Akashi et al.⁵ discussed that as a result of the injury of the gallbladder epithelium due to cholecystitis, the CA19-9 which is believed to be present in epithelial cells and in the bile inside the gallbladder will enter the blood stream. A similar mechanism is also presumed to be at work in xanthogranulomatous cholecystitis. Hong et al.⁶ previously discussed the proportion of cases with elevations in the levels of CA19-9 in xanthogranulomatous cholecystitis, and reported that since elevated levels of CA19-9 were found in 17 of 37 cases of xanthogranulomatous cholecystitis (46.0%), they concluded that the elevated levels of CA19-9 did not allow for distinction between xanthogranulomatous cholecystitis and gallbladder cancers. In addition, Zhuang et al.⁷ previously reported that the mean levels of CA19-9 in 45 patients with xanthogranulomatous cholecystitis and in

35 patients with gallbladder cancer were 602.8 ± 380.9 IU /l and 728.6 ± 261.1 IU /l, respectively; therefore, we should nonetheless note that the levels of CA19-9 can also be elevated in benign diseases such as xanthogranulomatous cholecystitis.

CONCLUSION:

CA19-9 is strongly expressed in malignant tumors of the digestive system and is widely used as a marker for gastrointestinal cancer. Xanthogranulomatous cholecystitis can also present with moderate elevation of CA19-9 levels, thus elevated CA19-9 cannot be used as a distinction between xanthogranulomatous cholecystitis and gallbladder cancers. Thus, biopsy and intraoperative frozen can help in its distinction and the patient can be managed accordingly.

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