Unusual association of diseases/symptoms

Heterogenous wall thickening of gall bladder: xanthogranulomatous cholecystitis or carcinoma, with type 3 choledochal cyst

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Summary

Xanthogranulomatous cholecystitis is an unusual inflammatory disease of the gallbladder characterised by severe proliferative fibrosis and the accumulation of lipid-laden macrophages in areas of destructive inflammation. Its macroscopic appearance may occasionally be confused with gallbladder carcinoma. The authors present a case of xanthogranulomatous cholecystitis with type 3 choledochal cyst in a 20-year-old man who was referred to our hospital with a 1-week history of abdominal pain and fever. He underwent endoscopic sphincterotomy and then open cholecystectomy. A histological diagnosis of xanthogranulomatous cholecystitis was made.

BACKGROUND

We consider this case as important because the presence of hypoechoic areas or cavities in a thickened gallbladder wall, together with calculi in a patient with chronic disease, is highly suggestive of xanthogranulomatous cholecystitis. The preoperative identification of xanthogranulomatous cholecystitis can be important in the proper surgical management of these patients and in the differential diagnosis with other gallbladder diseases.

CASE PRESENTATION

A 20-year-old male patient was admitted in our hospital with pain in right hypochondrial region, vomiting and fever for 1 week.

Fever was low grade and intermittent type. The patient also had increased pain following meals.

The patient had no history of any form of addiction and his family history was also insignificant.

INVESTIGATIONS

Apart from routine investigations which showed elevated white blood cell counts, the patient underwent abdominal sonography.

Ultrasonography revealed heterogenous thickening of wall of gallbladder with wall thickness measuring 20mm. There was evidence of increased vascularity within the wall of gallbladder with multiple cystic or hypoechoic areas. There was evidence of irregularity on luminal aspect of the gallbladder with presence of sludge and echoes within the gallbladder. Around 18 mm size single calculus, impacted at neck of the gallbladder was noted. Based on those features, diagnosis of xanthogranulomatous cholecystitis was made (figure 1). However, second possibility of carcinoma was also considered. Intrahepatic biliary radicles and common bile duct (CBD) were not dilated. Approximately 22×18 mm size cystic lesion was noted in relation to distal end of CBD within the duodenal lumen containing few hyperechoic foci casting acoustic shadowing suggestive

Figure 1 Ultrasonography image showing gall bladder filled with sludge and echoes with heterogeneous wall thickening containing multiple cystic areas within it and large calculus impacted near neck of gallbladder.

Figure 2 Ultrasonography image showing choledochal cyst and its relation to distal end of common bile duct with few calculi seen within it.

of stones with largest measuring 9 mm in size. The features were consistent with choledochal cyst type 3. Further investigations in form of CT, MRI and magnetic resonance cholangiopancreatography were advised (figure 2).

The patient did not undergo further investigations due to financial problem.

DIFFERENTIAL DIAGNOSIS

- ▶ Gallbladder carcinoma.
- ▶ Acute cholecystitis with perforation.

TREATMENT

The patient first underwent endoscopy (images not available) for management of type 3 choledochal cyst. Ultrasonographic findings were confirmed and sphincterotomy was performed and stones were removed. Stent was introduced in CBD.

On the following day, the patient underwent open cholecystectomy. Gallbladder was found adhered to adjacent omentum. Adhesiolysis was done and gallbladder was removed. Specimen was sent for histopathological examination.

OUTCOME AND FOLLOW-UP

Histopathological examination showed a mixture of ceroid (waxlike) xanthogranuloma with foamy histiocytes, multi-nucleated foreign body giant cells, lymphocytes and fibroblasts containing areas of necrosis. No evidence of malignancy was noted. The feature was suggestive of xanthogranulomatous cholecystitis (figure 3).

DISCUSSION

Xanthogranulomatous cholecystitis is an uncommon form of chronic cholecystitis, representing between 0.7% and 13.2% of gallbladder disease^{1 2} and mainly affecting women between 60 and 70 years old.^{1 3 4} Its importance lies in the fact that clinically and radiologically it can be confused with the carcinoma of the gallbladder.^{2 5}

Although the mechanism remains unclear, extravasation of bile into the gallbladder wall, with involvement of Rokitansky-Aschoff sinuses appears to be a precipitating factor.³ The presence of calculi or biliary tree obstruction may play an important role as in our case.⁶

The vomiting, upper right quadrant pain and leukocytosis observed in our patient is similar to the findings described in other types of cholecystitis. Of greater interest

Figure 3 Histopathological image showing typical foamy histiocytes, eccentric nuclei and abundant foamy cytoplasm characteristic of xanthogranulomatous cholecystitis.

is the possible association of xanthogranulomatous cholecystitis with other diseases, especially tumours of the gallbladder and bile ducts. Although disagreement exists on this matter,⁷ reported series support the existence of this association.^{2 8}

Although gallstones and a thickened and echogenic gallbladder wall are frequent radiologic findings, they are non-specific. The presence of sludge is also common.¹ Hypoechoic nodules and bands in the gallbladder wall are thought to be the most characteristic findings in the disease.^{1 3 9} As with sonography, thickening of the gallbladder wall with a hypoattenuated band around the gallbladder was also the most frequent CT finding.^{6 10} Usually there is homogeneous uptake of contrast material by the gallbladder mucosa. Role of MRI is controversial. Only one published report exists of MRI in a patient with xanthogranulomatous cholecystitis; those authors could not differentiate xanthogranulomatous cholecystitis from gallbladder carcinoma.¹¹

Treatment is generally surgical removal of gallbladder with either laproscopic or open approach.

Learning points

- It is quite difficult to differentiate xanthogranulomatous cholecystitis from gallbladder carcinoma on imaging. Accurate differentiation usually requires histopathological examination.
- Xanthogranulomatous cholecystitis is often associated with calculus disease of gallbladder.
- Type 3 choledochal cyst that was found in our case, may have contributory effect in pathogenesis of disease as it may cause obstruction or it may be a simple association.
- Though xanthogranulomatous cholecystitis is quite common in older age group (60s and 70s), it must be considered even in young individuals if imaging features are corresponding.

Competing interests None.

Patient consent Obtained.

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