Case Report



Laparoscopic Management of Unusual Presentation and Age of Xanthogranulomatous Cholecystitis, Case Report and Literature Review

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Abstract

Background: Xanthogranulomatous cholecystitis XGC is a rare variant of Cholecystitis characterized by extensive inflammation and Fibrosis and associated with more complicated presentation like invading other organs and fistula formation. it could mimic Underlying malignancy like Gallbladder Cancer GBC. Challenges started by difficulty of diagnosis pre operatively due to multi factors include less threshold of diagnosis, which can lead to over treatment if GBC suspected. Gallstones plays major role in pathogenesis of most variant of Cholecystitis and can be trigger for GBC. <u>Case presentation</u>: we report 2 cases with different pre-operative course, both cases managed with laparoscopic cholecystectomy, and Histopathology showed Xanthogranulomatous cholecystitis XGC. <u>Discussion</u>: recent cases reported for XCG usually occurs in 5th and 6th decades and presented with multiple presentation and could be missed diagnosis, laboratory and radiological investigations can offer a limited diagnosis if not interpreted by expertise. Management can vary from simple laparoscopic cholecystectomy -as in our cases – and can extend to major en block resection.

<u>Keywords:</u> Xanthogranulomatous cholecystitis, cholecystitis, gallbladder cancer, cholecystectomy, enhanced mucosal line.

Background

Xanthogranulomatous cholecystitis (XGC) is a rare inflammatory disease of the gallbladder characterized by severe proliferation of fibrotic tissue accompanied with the accumulation of lipid-laden macrophages and acute and chronic inflammatory cells ^[1,2]. Incidence of XCG reported between 1.3%-5.2% of resected Gallbladder specimens [1-3]. The most common chief complaints of the patient are Abdominal pain, others symptoms include Vomiting, nausea, changes in bowel habits ^[1,4] obstructive jaundice and its sequels can occur in about 20-25 % [2] weight loss and palpable mass also reported ^[2,4,5]. Ultrasound abdomen is considered first modality of radiological investigation and can showed thickness of gallbladder wall, intramural nodule, gallstone, or pericholecystic fluid ^[2,5,6]. CT scan plays the major role of Diagnosis in which the finding can distinguish the XGC from GBC that includes: diffuse or focal wall thickening, intramural hypoattenuating nodules in thickened walls, luminal surface enhancement with continuous mucosal lines or mucosal lines ^[1,2,6]. Management can vary from simple laparoscopic cholecystectomy as in our cases and can extend to major en block resection.

Cases presentation

Case I

History and case course: 35 years old Saudi female not known to have any medical illness, Patient present to the ED with 2 months history of RUQ abdominal pain and nausea and vomiting that increased last week. Initial investigation revealed diagnosis of Acute calcular cholecystitis. patient admitted and kept for conservative management on IV antibiotics until improved and discharged home. In the same admission obstructive jaundice suspected due to elevated Bilirubin so MRCP requested and the bile duct was clear and not dilated. Patient assessed in the OPD and booked for surgery after 8 weeks of the attack.

Patient re admitted electively for Laparoscopic cholecystectomy as Day surgery case, preoperative measurement taken and laparoscopic access achieved. Intraoperative finding : severe adhesions of the gallbladder to the omnetum and adjacent structures , trial of adhesionlysis by the electrocautery hook and Maryland's instruments until the GB completely released, critical view of safety achieved and cystic duct and artery clipped and ligated , release of Gallbladder from liver bed took time due to severe adhesion of GB to liver bed in which part from posterior wall left on place , hemostasis achieved specimen removed and closure as usual laparoscopic case. Patient discharge at the late evening with good condition. Follow up in the clinic patient

completely fine and clips removed. Histopathology sowed XGC Xanthogranulomatous cholecystitis

Investigations

U/S showed the gall bladder is normal in size, shape and increase in wall thickness. With multiple variable sized intraluminal stones averaging 7mm. (Figure 1)

MRCP showed Distended gall bladder with pericholecystic fluids with multiple gallstones. Gall bladder measuring 11x5 cm. Gall bladder neck stone is noted measuring 1 cm. The common bile duct normal in diameter. No CBD stones. (Figure 2)

Case II

History and case course: 28 years old Saudi male not known to have any medical illness, Patient present to the clinic with frequent RUQ abdominal pain, nausea and vomiting On-off course, increased with fatty meals. Clinical examination unremarkable. Preoperative investigation revealed gallstones. Patient admitted electively for Laparoscopic cholecystectomy as Day surgery case. preoperative measurement taken and laparoscopic access achieved. Intraoperative finding: similar to Case I, severe omental adhesions and duodenal adhesions. Trial if adhesinolysis by blunt dissection and by electrocautery using Hook and Maryland's instruments. Gallbladder released from the adhesions and critical view of safety achieved with no difficulties, cystic duct and artery clipped and ligated safely. Releasing the GB from liver bed was challenging due to loss of the hepatocytic plane. GB removed and hemostasis achieved and laparoscopic closure as usual. Post op period was uneventful and patient discharge late evening of the same day. Follow up in the clinic was unremarkable clips removed, Histopathology and all showed Xanthogranulomatous cholecystitis (Figure 3)



bladder with pericholecystic fluids with multiple gallstones



Figure (3) Post operative Histopathology slide with Feature of XGC include foamy macrophages with mucosal lining and inflammatory cells infiltration

Discussion

Preoperative diagnosis of XGC considered a challenging step for the surgeon due to several reasons. First, it has similar clinical features of any other gallbladder diseases like Biliary colic, acute or chronic cholecystitis. Second, not all of radiological features of the diseases can be present in the preoperative studies. Third, due to its rare incidence, the suspicion of diagnosis is low between the physicians and radiologists.

Laparoscopic Cholecystectomy considered as the gold standard procedure for all benign gallbladder diseases. Role of Laparoscopic approach is still the best option for management of XGC. The contraindications of laparoscopic cholecystectomy include suspicion of malignancy on the pre operative images, intraoperative severe adhesion that could lead to bile duct injuries (BDI) or adjacent structures injuries. Expert Minimal invasive or hepatobiliary surgeon is better for management of such these cases. In the mass forming variety of XGC the intraoperative decision can extend to radical and multi staging surgeries ^[5].

XGC still not proven as premalignant lesion ^[4,5]. Although they can present together but the pathogenetic link between them isn't proven yet. As Gallstone considered as Risk factor for both the theory support that it can be 2 pathological processes at the same time.

Other modality of diagnosis added in the literatures to aid in the preoperative diagnosis. MRI abdomen with Biliary tree MRCP doesn't offer any advantages over Contrasted CT scan ^[5]. PET scan also been used to differentiate between XGC from GBC with reporting of false positive and false negative which decrease its sensitivity and specificity ^[5]. Tumour markers are used for specific tumour producing antigens like CEA and Ca19.9 in both cases should elevation which make the distinguish between the two disease is difficult ^[1,5]. Ultrasound-guided fine needle aspiration cytology (FNAC) is used in some centres with high failure rate of diagnosis ^[1,5].

Post-operative complications like any other abdominal surgery and differ in incidence and severity from laparoscopic and open surgery. Wound Infections, bile fistula, cholangitis, and residual choledocholithiasis ^[4]. Rate of conversion to open cholecystectomy is higher in XGC than other forms of cholecystitis ^[1]. There was wide variation in the proportion of patients for whom treatment was attempted laparoscopically, being lowest in India and the Americas and highest in Europe and the Far East ^[6].

Learning Points

- Xanthogranulomatous cholecystitis is difficult diagnosis and most of cases discovered intraoperatively which need to increased suspicion of the diagnosis if adhesions and chronic inflammatory process present.
- Focusing in pre-operative radiological studies can mimic radical procedure if cancer suspected falsely.
- Laparoscopic approach is still the gold standard of care in gallbladder disorder except of malignancy suspected or laparoscopic approach can cause bile duct injuries or adjacent organ injuries.
- Laparoscopic or hepatobiliary surgeons are the preferable to manage and treat such of these cases if discovered preoperatively.

Ethics approval and consent to participate

Not applicable

List of appreciations

XGC: Xanthogranulomatous cholecystitis. GBC: Gallbladder Cancer.

Conflict of interest

All the authors declare that there is no conflict of interest for the publication of this paper.

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