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Case report

A rare case of xanthogranulomatous cholecystitis mimicking gall bladder carcinoma

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ABSTRACT

Xanthogranulomatous cholecystitis is an unusual form of benign chronic cholecystitis with focal or diffuse destructive inflammatory process characterized by thickened fibrotic disrupted gall bladder wall with foamy histiocytes and bile extravasation. Its significance lies in the fact that it may simulate malignancy clinically, radiologically, pathologically and on the top of that it may coexist with gall bladder carcinoma leading to diagnostic dilemma.

1. Introduction

We hereby present a case of 60 year old male who was admitted in our surgical unit with complaints of pain right upper quadrant of abdomen and yellowish discoloration of urine for 3 months but with normal colour of stools. His vital parameters were normal. On abdominal examination, guarding and tenderness was present in right upper quadrant but no abdominal lump was present. His laboratory investigations were Hb 12gm%, TLC 8,000, total bilirubin 3.4mg/dl(conjugated 1.4, unconjugated 2mg/dl), AST 101 IU/l, ALT 47 IU/l, ALP 181 IU/l. USG whole abdomen showed edematous gall bladder wall with multiple gall stones with rest of the solid organs being normal. His CECT abdomen showed diffuse asymmetrical thickening of gall bladder wall predominantly involving the fundus with mild diffuse enhancement with cholelithiasis with necrotic lymph nodes at porta hepatis (**figure 1 and figure 2**), suggesting features of carcinoma Gall bladder. After proper pre-operative anaesthatic checkup, pre-operative antibiotics and hydration, patient was subjected to extended cholecystectomy. Per-operatively gall bladder was found to be contracted, thickened with adhered omentum. The cut section showed lumen filled with black colored pigment stones and sludge with gall bladder wall showing yellowish areas but no well defined growth. On microscopic examination, mucosa exhibited marked intestinal metaplasia with focal dysplasia with mucosal glands seen reaching deep down to serosa. Gall bladder wall showed multiple foamy macrophages and multinucleate giant cells with chronic inflammatory cell infiltrate (**figure 3 and figure 4**). The perihepatic and pericholedochal lymph node sections showed non-specific reactive hyperplasia. Sections from gall bladder bed showed marked xanthogranulomatous inflammation and chronic inflammatory cell infiltration and hemorrhage. A provisional diagnosis of xanthogranulomatous calculus cholecystitis was made. Postoperative period was uneventful and patient was discharged on 10th postoperative day in satisfactory condition.

2. Discussion

Xanthogranulomatous cholecystitis (XGC) is an idiopathic rare inflammatory lesion of the gallbladder characterized by focal or diffuse destructive inflammatory process with marked proliferative fibrosis, infiltration of macrophages and foam cells involving the wall of the gallbladder with gallbladder inflammation, pericholecystic infiltration, hepatic involvement and lymphadenopathy which may present as mass lesion with adjacent organ invasion like carcinoma gall bladder[1,2]. Similar inflammation may occur in kidneys, skin, retroperitoneum, genitourinary tracts and cranial cavity. This entity was first described by Christensen and Ishak in 1970 who called it pseudotumour of gallbladder and it was named xanthogranulomatous cholecystitis by Mccoy in 1976[3,4]. XGC is found after approximately 0.5%-1.8% of routine cholecystectomies almost always in presence of gallstones (91%-100%)[5,6]. The incidence of XGC is reported to be 0.7% to 13.2%[1,7] and its highest incidence has been found in our country[8].

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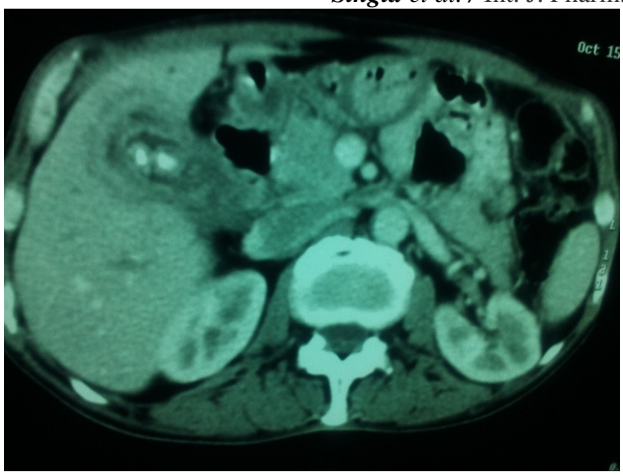

Figure 1

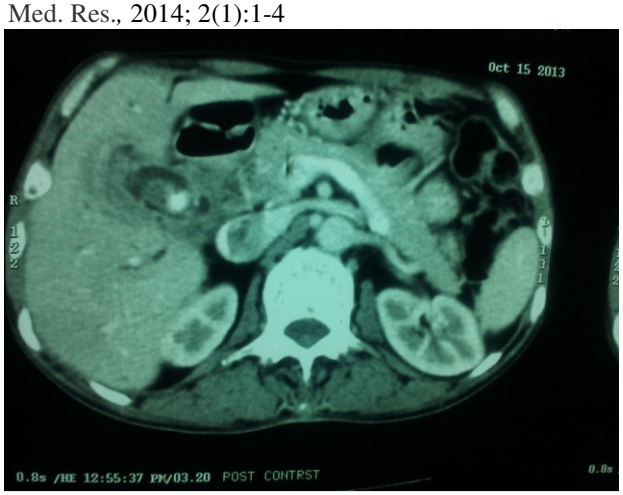

Figure 2

Figure: 1 &2 Shows CECT abdomen showed diffuse asymmetrical thickening of gall bladder wall predominantly involving the fundus with mild diffuse enhancement with cholelithiasis with necrotic lymph nodes at porta hepatis)

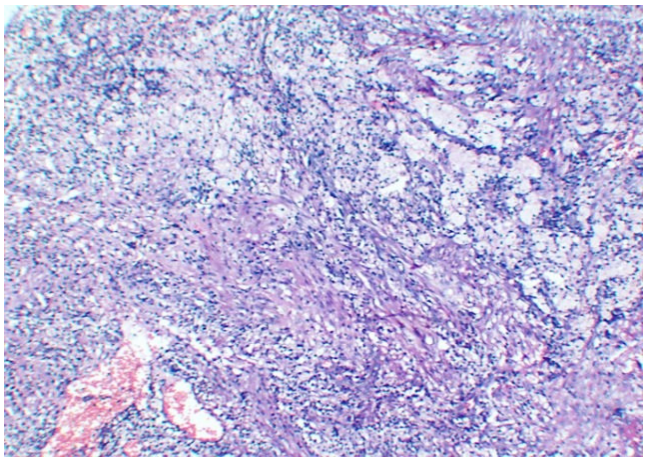

Figure 3: H & E 40X Sheet of foamy cells with chronic inflammatory cell infilterate

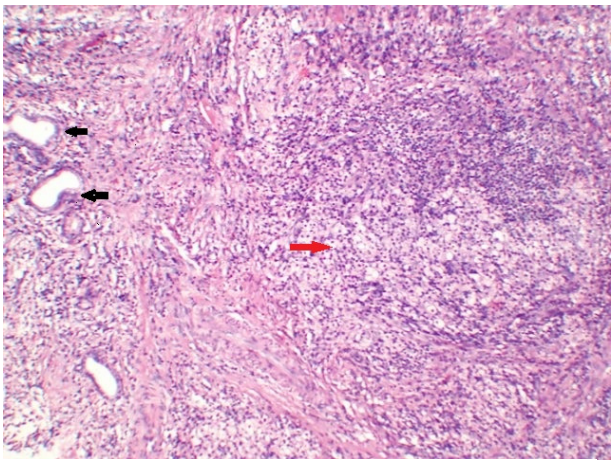

Figure 4: H&E 10X Gall bladder wall exhibiting mucosal gland (black arrows) and sheet of foamy cells with chronic inflammatory cell infiltrate (red arrow)

It most commonly occurs in middle aged women and 6th to 7th decade of life in males with slight female preponderance and M:F ratio has been found to be 2.6:1[9-13]. In our country mean age has been found to be 48.2 years[14]. Its etiology include large gallstone volume with hypo-contractile gall bladder and are present in most patients[15-17]. Its pathogenesis includes rupture of Rokitensky-Aschoff sinuses and extravasation of bile in muscular layer in presence of gallstones, obstruction and cholestasis which leads to formation of lysolecithin. This leads to further damage to gall bladder mucosa which leads to infilteration by inflammatory cells with resultant phagocytosis of bile pigments hemosiderin and cholesterol giving the typical picture of xanthogranulomatous cholecystitis with presence of foamy macrophages with acute and chronic inflammatory cells[18,19]. Grossly, gall bladder may be enlarged or fibrosed with its mucosa being ulcerated or atrophic and thickened wall with formation of yellowish nodules. Microscopically there is presence of inflammatory cells, lipid laden macrophages and foreign body giant cells with severe proliferative fibrosis but it lacks true malignant features like pleomorphism, cellularatypia or mitotic figures. As the disease progresses there is fibrous hyperplasia and inflammatory granuloma formation which in turn leads to gall bladder thickening and adhesion to adjacent structures[20]. Recently Zhuang et al. have found that xanthogranulomatous cholecystitis contains upregulated oncogenes like BCL-2 and c-MYC suggesting its precancerous nature but needs further research in this direction[21]. The clinical and laboratory findings of cases with Xanthogranulomatous cholecystitis are nonspecific and similar to those of acute or chronic cholecystitis[22]. Clinically patient may present with right sided abdominal pain, nausea, vomiting and jaundice. Patients with malignancy are more likely to present with anorexia, weight loss, palpable mass and jaundice. IN XGC, ultrasound can detect focal or diffuse thickening of the hyperechoic gallbladder wall with the presence of characteristic intraluminal hypoechoic nodules in 35-73% of

the cases[22,12]. Other commonly reported findings are cholecystitis-like fluid collections near the gallbladder and dilation of intra- and extra hepatic bile ducts in case of choledocolithiasis '.B-mode USG reveals hypoechoic nodules and low level echo band[12]. CT scan findings of xanthogranulomatous cholecystitis reveals intramural irregular hypo attenuated soft tissue mass with mucosal line in thickened lobulated gall bladder[1,12,24,25]. MDCT has proved to be better than conventional CT because it helps to assess enhancement pattern by allowing dynamic images, improved spatial resolution with thinner sections, and provides the multiplanar reconstruction images in addition to the axial image[25,26].It has been found that diffusion weighted magnetic resonance imaging (DWI) may be most useful imaging modality in differentiation of xanthogranulomatous cholecystitis from gall bladder carcinoma as its sensitivity and specificity are 79% - 86% and 94.7% respectively[28]. 18f-FDG PET has also been reported to be very useful in differential diagnosis of gall bladder carcinoma with its sensitivity being 75.5-78% and specificity being 82%-100%[29,30,31]. Imaging modalities like ERCP, PTC have also been found to be useful especially if common bile duct involvement is suspected but despite all above imaging modalities definitive diagnosis is made only histologically. Preoperative fine needle aspiration biopsy and intraoperative frozen section have also been found to be valuable in differential diagnosis when there is no invasion of adjacent organs.

Subtotal or Extended cholecystectomy with excision of the whole surrounding xanthogranulomatous tissue has been advocated to be standard approach to prevent future recurrence[1,11,32]. Due to dense adhesions in and around Calot's triangle, laparoscopic cholecystectomy is difficult to be performed and associated with higher risk of complications. In cases where malignancy cannot be ruled out even after a complete preoperative diagnostic examination and there is presence of symptoms such as jaundice and cholangitis, as well as the potential risk of life -threatening complications like bowel obstruction and subsequent perforation, radical surgical approach is strongly recommended. The intraoperative and postoperative morbidity rates are higher in xanthogranulomatous cholecystitis and it is associated with various complications like hepatic abscess formation, perforated gall bladder, cholangitic stenosis, entero-biliary fistula, billiary peritonitis, wound infection, pleural effusion, acute renal failure, etc .

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