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Cystic Artery Pseudoaneurysm Secondary to Xanthogranulomatous Cholecystitis: A Rare Manifestation

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Abstract: ***Background: Pseudoaneurysm of cystic artery is very uncommon in the setting of chronic Xanthogranulomatous cholecystitis. When these pseudoaneurysms rupture they can lead to dreaded complications like hemoperitoneum. Radiopathological findings and surgical approach have been explained in our case. As patient was hemodynamically stable, no further invasive procedures were performed. Laparoscopic cholecystectomy was felt as a safe option.Histological examination of gall bladder confirmed radiological finding of Xanthogranulomatous type of chronic cholecystitis***

Keywords: Xanthogranulomatous, Hypodense nodules, Pseudoaneurysm, Laparoscopic

1. Case Report

A 73-year-old man was referred for an ultrasound of the upper abdomen with a complaint of pain in right hypochondrium since 1month. No history of jaundice, vomiting and fever noted. Ultrasound scan revealed a large calculus measuring 1.8cm in the fundal region. The gallbladder had thickened walls (13.2mm shown in fig I) and was inpartially distended state. A rounded cystic structure was present within the neck of the gallbladder. This structure demonstrated swirling(ying yang) flow signals on colour Doppler study. There was no pericholecystic fluid or ascites. The appearances on the ultrasound and colour Doppler scans were considered to be indicative of pseudo-aneurysm.

CT scans of the abdomen were performed with intravenous contrast in the arterial and venous phases for confirmation of USG finding of a pseudo-aneurysm in the neck .Gall bladder showed loss of fat planes with the adjacent part of liver. A large laminated hypodense calculus measuring 17.2x19mm was seen in the fundal region in intramural location with focal mucosal ulceration(Fig III).Multiple hypo dense nodules were noted in the diffusely thickened wall. There was a well-defined partially thrombosed aneurysm arising from the cystic artery in the wall of gall bladder neck bulging into the lumen(Fig II). The opacified lumen measures 11.9mm x 10mm.The thickened gall bladder were seen abutting the duodenum without any e/o wall thickening. CBD was normal. No IHBR dilatation seen

Following the CT scan, the patient was admitted to hospital. On admission, his vitals were stable. His haemoglobin was 10 gm%, White cell count was 7.9x1000/cummwith 74.7% neutrophils and platelets were 213& × 1000/cumm. Coagulation profile was normal.

He was discharged with a plan for laparoscopic cholecystectomy with pseudoaneurysm excision which was performed within a 15 days. Trans catheter embolization was not done as there was no evidence of rupture and patient was hemodynamically stable. Pneumoperitoneum was created through umbilicus and the standard 4-port laparoscopic cholecystectomy approach utilised. Superficial branch of cystic artery was involved (Fig IV)Cystic artery and duct were clipped and gall bladder was removed safely. The histopathology report on the gallbladder specimen showed chronic inflammatory cell infiltrate along with foamy histiocytes. No malignancy was seen. The histopathological findings are consistent with Xanthogranulomatous cholecystitis.

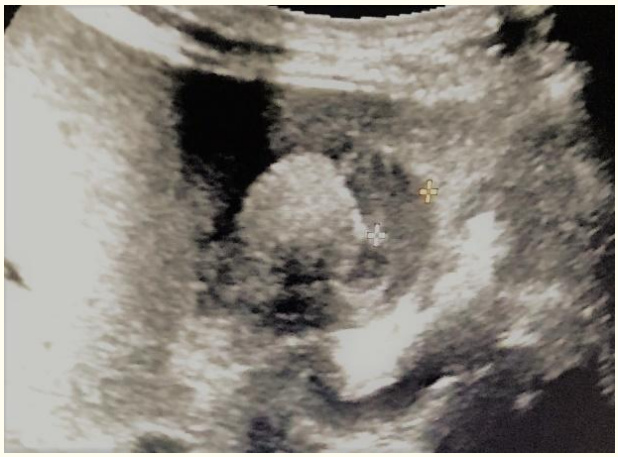

Figure 1: USG shows diffuse gall bladder symmetrical wall thickening in the fundal region

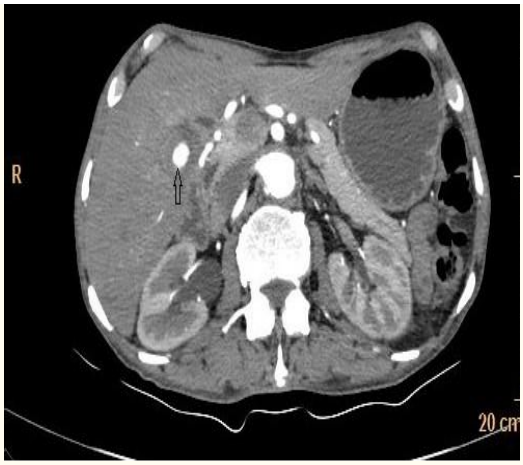

Figure 2: Axial contrast enhanced CT of the abdomen shows a well-defined enhancing round pseudoaneurysm of cystic artery in the region of neck of gall bladder with eccentric thrombosis.

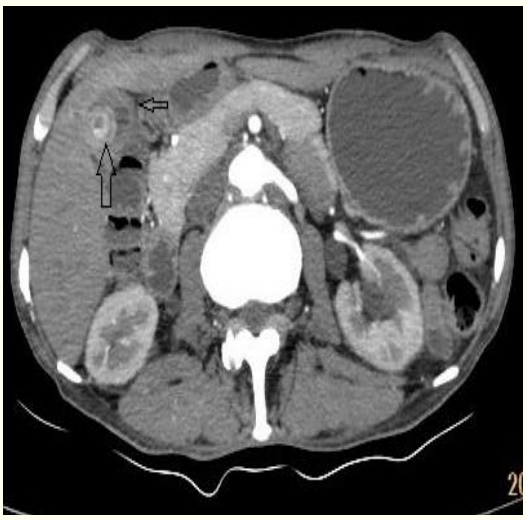

Figure 3: Axial contrast enhanced CT image shows a lamellated calculus in the fundal region(long arrow) and diffusely thickened walls with intramural hypodense nodules and ulcerations(small arrow)

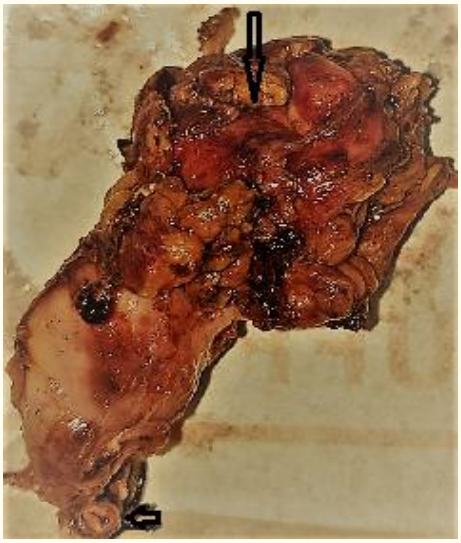

Figure 4: Gross specimen shows multiple yellowish xanthoma nodules within the diffusely thickened walls (long arrow) with cystic artery pseudoaneurysm in the neck region (small arrow).

2. Introduction

Cystic artery pseudoaneurysm is a very uncommon entity.[1]The maximum no. of cases seen are complications of biliary and angiographic procedures due to breech in the arterial wall Pseudoaneurysms secondary to chronic calculus Xanthogranulomatous cholecystitis are seen rarely.[1] Patients typically present with symptoms and signs similar to cholecystitis including right upper quadrant pain with a positive murphy sign.[2]Male preponderance has been reported with a male to female ratio of 2:1[3]

The pathophysiology of aneurysmal dilatation of the cystic artery is thought that the artery is eroded either by direct pressure of gallstones or inflammation of the arterial wall. Xanthogranulomatous variety is characterised by presence of multiple intramural nodules. This consequently leads to damage of the adventitia with localized weakness in the adjacent vessel wall and formation of a pseudo aneurysm.[4] Early management is essential because rupture of aneurysm can lead to hemobilia and, upper GI bleed and hemoperitoneum.[5]

We hereby describe our case of an unruptured cystic artery aneurysm in the context of chroniccalculus xanthogranulomatous cholecystitis, which was detected on CT and confirmed on histopathological examination of gall bladder removed laparoscopically.

Discussion

Xanthogranulomatous cholecystitis is a rare prototype of chronic cholecystitis characterised by diffuse ongoing inflammatory mechanism followed by infilteration of lymphocytes and foam cells[6]. Complications are seen in 32% of patients which include abscess formation , fistula and rarely a pseudoaneurysm. Proposed theory behind Xanthogranulomatous etiology is ulceration of mucosa or disruption of Rokitansky –Aschoff sinuses due to elevated intraluminal pressure secondary to calculus which causes seepage of bile in the walls of gall bladder.[3-6] The intramural nodules detected on imaging studies (85.7% and 61.1 % by Zhao et al[7]The association of Xanthogranulomatous cholecystitis with gall stones are seen in 80% of the patients.[8]According to literature only approx.16 cases reported an unruptured cystic artery pseudoaneurysm secondary to Xanthogranulomatous etiology with median age of presentation 65 years making our case likely 17th in the literature. Some of them were managed with open cholecystectomy and ligation of the cystic artery [9] while the others were managed laparoscopically .[10] In high risk patients with active bleeding angiographic embolisation has been done [11] In our case we have described that laparoscopic approach of gall bladder removal and cystic artery pseudoaneurysm an appropriate and safe option.

3. Conclusion

Xanthogranulomatous cholecystitis can be a diagnostic dilemma and a correct preoperative diagnosis can be aided by knowledge of characteristic findings on CT and MRI with histopathogical correlation. These unruptured pseudo-

aneurysms may be safely treated with laparoscopic approach in the hands of an experienced surgeon.

Conflict of interest: None

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