

Diagnostic Shift from Gall Bladder Carcinoma to Periapillary Tumor Owing to Xanthogranulomatous Cholecystitis: A Narrow Escape

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Abstract

Xanthogranulomatous Cholecystitis (XGC) masquerading as gallbladder cancer is not uncommon. Imaging modalities have their own limitations in the diagnosis of XGC and the final confirming diagnosis is by histopathology. Unnecessary radical resections, increased cost, higher morbidity and mortality rates are also associated with XGC. We present a case of obstructive jaundice primarily due to periapillary tumor but complicated by XGC, creating a diagnostic dilemma and shifting the focus to gall bladder carcinoma (GBC) preoperatively. The index case is about how this mystery was resolved and treated successfully.

Keywords: *Xanthogranulomatous cholecystitis; Gall bladder carcinoma; Periapillary tumor*

1. Introduction

Xanthogranulomatous cholecystitis (XGC) is an inflammatory disease and uncommon variant of chronic cholecystitis [1]. Focal or diffuse destructive inflammatory process followed by marked proliferative fibrosis along with infiltration of macrophages and foamy histiocytes (xanthoma cells) are the main characteristic features of XGC [1]. The incidence of XGC is variable and has been described between 0.6% to 10% [2]. There are various reports of Xanthogranulomatous Cholecystitis

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masquerading as gallbladder cancer. Although different imaging modalities have their role in the diagnosis of XGC but frequent misdiagnosis by the different imaging modalities is not uncommon [3,4]. The conclusive diagnosis is by histopathology [5-8]. Given the relative scarcity of the disease and the fact that it may be difficult to differentiate from gallbladder carcinoma (GBC) based on clinical presentation and preoperative imaging, it is not uncommon that patients with XGC are taken to the operating room without a clear diagnosis. Our index case is an example of diagnostic dilemma owing to Xanthogranulomatous Cholecystitis where the primary was periampullary tumor in an obstructive jaundice patient, but the focus was on the gall bladder. However, this diagnostic uncertainty successfully resolved and treated.

2. Case Presentation

A 65-year, gentleman, presented with high coloured urine and yellowish discolouration of eyes for the last 1 month to outpatient clinic. There were no known comorbidities. He had loss of appetite but no significant loss of weight yet. On evaluation, he was found to have features of obstructive jaundice and elevated CA 19.9 (1068 U/ml). He underwent MRCP which showed cholelithiasis, dilated CBD with? stricture at lower end. EUS with side view endoscopy was done which showed normal liver, gross intrahepatic biliary radicles dilatation, dilated CBD with distal abrupt cut off. GB was distended with a calculus. There was irregular thickening of the GB wall. The final impression was EHBO- distal CBD stricture? gall bladder carcinoma. Endoscopic biopsy from the periampullary region, done in view of bulky papilla with slight irregularity in the mucosa, did not show any malignancy. Patient's bilirubin (Total Bilirubin -16 mg/dl, Conjugated Bilirubin 8.7 mg/dl) was on increasing trend when he was subjected to ERCP and stenting. Bilirubin and CA 19.9 were started coming down, but diagnosis was still a challenge. A multiphase CECT was done to aid in the diagnostic process which further complicated the scenario by reporting 2 arterial enhancing sub centimetric lesions in segment 2 and 6 of the liver as? metastasis from carcinoma gall bladder. Gall bladder showed mildly thickened shaggy walls, 3 mm - 3.5 mm in fundus/mid body and 6 mm - 7 mm in the neck region (FIG. 1 & 2).

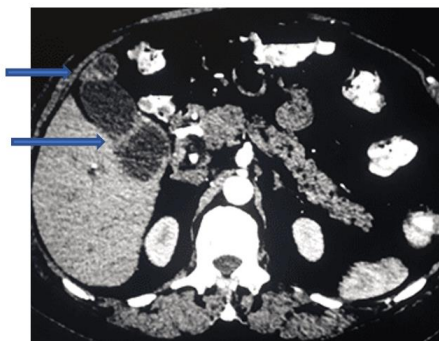


FIG. 1

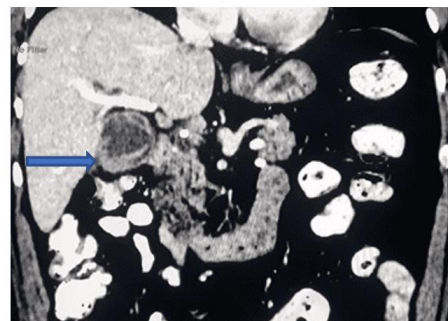


FIG. 2

FIG. 1,2. Triphasic CECT images showing irregular wall thickening in the fundus, body and neck region.

Following this, patient underwent PET scan which showed FDG avid gall bladder fundus wall with SUV max 3.9 and a 3.9 cm × 3.8 cm metabolically active (SUV max 6.7) cystic/necrotic lesion in relation to segment 6 of liver? liver lesion with exophytic component? lymph node with liver infiltration (FIG. 3,4). Distal CBD thickening with mild FDG uptake (SUV max 2.5) was seen in the periampullary region causing abrupt cut off. There were few periportal, portocaval and peripancreatic LNs, suspicious for metastatic disease. This really confused the matter and there was no clue to the primary diagnosis. But still the

focus was on carcinoma gall bladder as every imaging modality was favouring it. Meanwhile, the patient was very stable with increasing clinical status and decreasing bilirubin every day. The case was discussed in a multidisciplinary team and decided to do repeat biopsy from the periampullary region. The arrow hit the target this time and revealed well differentiated adenocarcinoma. Patient's Bilirubin and CA 19.9 were also got normalized by this time and he was planned for Pancreaticoduodenectomy with frozen section from gall bladder.

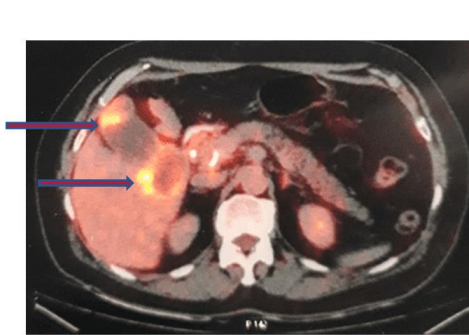


FIG. 3

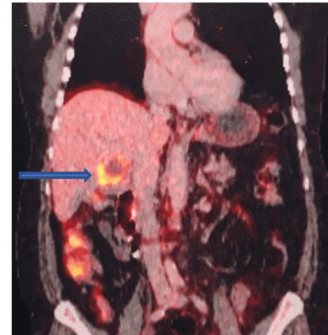


FIG. 4

FIG. 3,4. PET scan images showing FDG uptake in the GB- body and fundus, CBD stent also seen.

Intraoperatively, liver appeared normal with no space occupying lesions. There were no peritoneal, pelvic, mesenteric, or omental deposits and no free fluid. Gall bladder was diffusely thickened, covered by omentum and colon. Intraoperative frozen section from the gall bladder was negative for malignancy. Emyema of the gall bladder came into picture on sudden rupture and spill of the contents during a difficult cholecystectomy. There was a single stone of size 2 cm in the gall bladder lumen. As such there was no growth on digital palpation in the periampullary region. Patient underwent a classical pancreaticoduodenectomy. On gross examination there was a periampullary growth of 15 mm x 15 mm (FIG. 5). As the patient was having stent in the CBD and empyema of the gall bladder, considering the fact of post operative surgical site infection, the wound was intermittently tagged with Ethilon sutures followed by twice daily dressing in the post operative period. There was biochemical pancreatic fistula on post operative day 3 which resolved by day 5. Patient was started on FJ feed 1st followed by oral liquid and semisolids thereafter. There was not much of the component of delayed gastric emptying although he had nausea sometime and vomited once on post operative day 5 but there was never a requirement of re-insertion of Ryle's tube. Secondary closure of the wound was done on post operative day 6, a day prior to discharge.

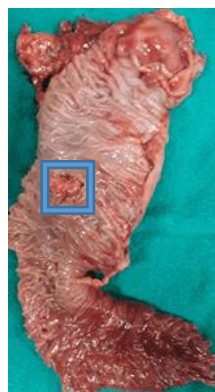


FIG. 5. Cut open gross specimen showing 15 mm x 15 mm tumor at periampullary region.

Histopathology reported as moderately differentiated adenocarcinoma of periampullary region infiltrating muscularis propria of duodenum (FIG. 6). All margins were free of tumor. One peripancreatic lymph node was positive with extra nodal extension turning the patient a candidate for adjuvant chemotherapy (FIG. 7). Surprising to us, gall bladder showed xanthogranulomatous cholecystitis, realizing the fact of inclination towards carcinoma gall bladder in the pre-operative period (FIG. 8). The diagnostic dilemma encountered during pre-operative period could be clearly attributed to xanthogranulomatous cholecystitis now. One could imagine doing radical cholecystectomy for xanthogranulomatous cholecystitis in view of carcinoma gall bladder and leaving the primary periampullary tumor intact, hence the title, “a narrow escape”. Patient received adjuvant chemotherapy and was doing well till last follow up.

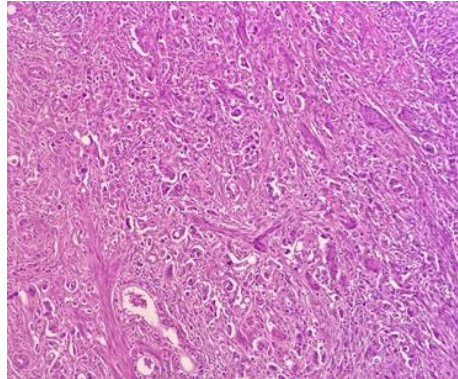


FIG. 6. Moderately differentiated adenocarcinoma periampullary region, 100×.

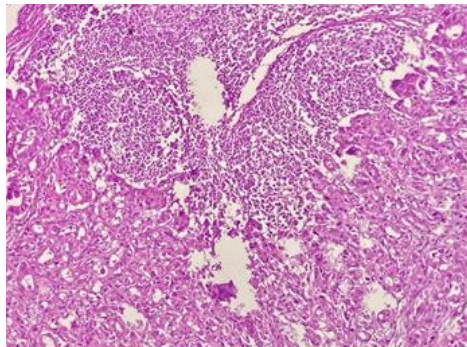


FIG. 7. Metastasis in peripancreatic lymph node, 100×.

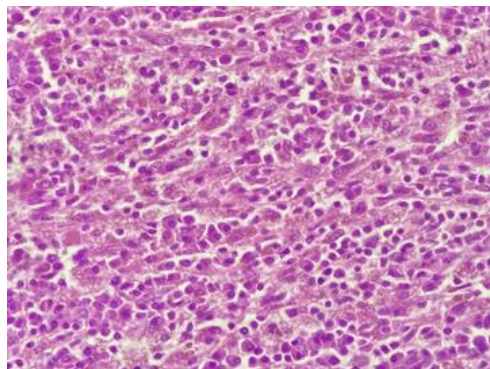


FIG. 8. Xanthogranulomatous cholecystitis, 400×.

3. Discussion

XGC is characterized by a diffuse, destructive inflammatory process with the gall bladder wall being infiltrated by foam cells and macrophages causing a proliferative fibrosis. Because it closely resembles GBC with pericholecystic infiltration, hepatic involvement and lymphadenopathy, it was termed as 'pseudo tumour' but later called xanthogranulomatous gall bladder [9,10]. Unnecessary radical resections or routine frozen section analysis in doubtful cases were being done for XGC because of its similar features with GBC [11]. Therefore, a preoperative diagnosis of XGC is important to avoid more extensive operations in patients with a benign disease. History of anorexia and weight loss favours GBC while fever and recurrent abdominal pain with a longer average duration points towards diagnosis of XGC [12]. The association of XGC with GBC is still a matter of debate [13]. EUS-guided FNA is a useful modality for sampling various targets. While a positive FNAC confirms the diagnosis of GBC, a negative sample does not rule it out. Intraoperative frozen section examination is an efficient method for exclusion of GBC [14,15].

In our case also, diagnostic uncertainty was the major challenge for appropriate management. The periampullary tumor was small in size which went unnoticed in almost every investigation and gall bladder pathology was obvious to catch everyone's eyes. Decrease in bilirubin level was expected post CBD stenting but on the other way, complete normalization of CA 19.9 was astonishing. Decision of the multidisciplinary team meeting to sample the periampullary region again owing to side view endoscopic findings and negative diagnosis on earlier biopsy led to the clueless and unexpected diagnosis.

4. Learning Points

The differentiation of XGC and GBC preoperatively and intraoperatively remains a challenge and a definitive diagnosis still needs a histopathological examination. An accurate preoperative diagnosis requires an integrated review of clinical and characteristic radiological features. The hidden malignancy in the shade of XGC should be strongly suspected as in the index case. Finally, the role of multidisciplinary team meeting is never forgettable.

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