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A RARE CASE OF HYALINISING CHOLECYSTITIS IN A PATIENT WITH GALLBLADDER CALCULI

General Surgery

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ABSTRACT

Hyalinizing cholecystitis is a rare variant of chronic cholecystitis in which the gallbladder tissue is replaced with hyaline sclerosis, more or less, and has a typical intra-operative appearance. Preoperative diagnosis may be difficult in the absence of suggestive features. It is strongly associated with gallbladder carcinoma. Therefore, the pathologist should perform a thorough microscopic analysis. We present a case of an unusual intra-operative finding in a female patient with chronic cholecystitis which was proved to be a hyalinising cholecystitis without carcinomatous changes.

KEYWORDS

gallbladder; calculi; chronic cholecystitis; hyalinising cholecystitis

INTRODUCTION

Chronic cholecystitis is a very common disease in North Indian population specially in females. Hyalinizing cholecystitis (HC) is a rare clinico-pathological subtype of chronic cholecystitis with hyaline sclerosis of the gallbladder wall without complete calcification as in the case of 'porcelain gallbladder'. Its incidence is ~1.6% of the cases of cholecystitis. A strong association of HC with gallbladder carcinoma has been reported. The frequency of gallbladder carcinoma arising in these patients is estimated to be 15% [1]. We are reporting a case of a female patient operated for symptomatic chronic cholecystitis, preoperatively diagnosed with ultrasound with no evidence of HC or carcinomatous changes. The intraoperative findings of unusual gall bladder appearance was later confirmed to be a case of HC. Informed consent was taken from the patient to present this case.

Case Report

A 50-year-old female patient was admitted to our hospital for an elective laparoscopic cholecystectomy due to chronic calculous cholecystitis. She had complaints of right upper abdominal pain with vomiting for 2 months. Pre-operative ultrasound suggested distended gall-bladder with multiple calculi. The gall-bladder wall appeared mildly thickened. Preoperative laboratory work-up showed no abnormalities except hypothyroidism.

Intra-operatively, a cystic structure, approximately 1.5X1.5cms, was identified near the gall-bladder fundus, which was densely adhered to the liver and posterior abdominal wall. Bladder wall was covered with whitish tissue replacing the normal gallbladder serosa (Fig. 1)

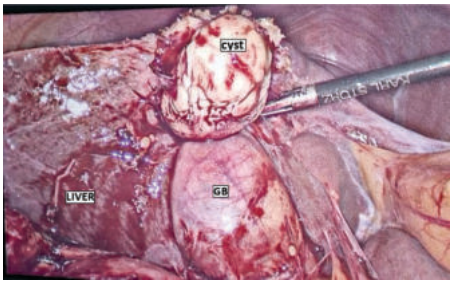

Fig.1 : Laparoscopic image of cyst arising from gallbladder fundus, densely adhered to liver and posterior abdominal wall.

No other abnormalities were noted during the exploration and after careful dissection, cholecystectomy was performed without intra-operative incident. Precautions were taken to remove the cyst, without rupturing it. In view of the meticulous dissection done at the operative site, an intra-abdominal drain was left in situ to avoid any collection.

The postoperative period was uneventful and the patient was discharged with abdominal drain at postoperative Day 2, to be seen again after a week. At Day 7, drain contained 10cc of sero-sanguinous collection, hence was removed. Histo-pathological analysis of specimen reported a chronic calculous hyalinising cholecystitis. On gross examination, gallbladder specimen was 6 cms in length. Outer surface was covered by fat. Mucosa was partially greenish with yellow streaks. Fundal muscosa was rough and pale white. Wall was thick, firm and calcified at fundus. Cystic duct was identified. On microscopic examination, mucosa showed inflammatory changes with aggregates of lymphocytes, plasma cells admixed with eosinophils and foamy histiocytes. Wall revealed muscular hypertrophy with transmural inflammation, fibrosis and congested blood vessels. Fundus showed paucicellular hyaline sclerosis with focal calcification (Fig. 2). There was no evidence of dysplasia or malignancy in any of the sections examined.

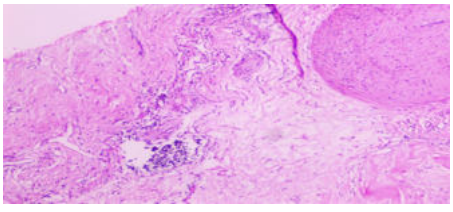

Fig. 2 : Histopathology slide showing hyalinizing sclerosis of gallbladder fundus.

DISCUSSION

HC was initially described by Patel et al. as a distinct type of cholecystitis in 2011. They described the replacement of the histological gallbladder layers with dense, paucicellular hyaline fibrosis with or without calcifications [1, 2]. The gender distribution for HC has shown a female predilection with reported female-to-male ratio of 5:1.Mean patient age being 56 years [1]. The histopathological definition of the HC suggests the presence of dense, laminar, paucicellular, or acellular hyaline fibrosis of the gallbladder wall, forming a thin and uniform band. Another distinctive feature of the HC is the presence of calcifications. Cases with diffuse, morethan 80% intramural calcifications are called 'porcelain gallbladder'. Finally, HC may also be associated with adenocarcinoma [1, 3]. In the present case, focal fundal calcifications were noted but there was no evidence of dysplasia or carcinoma in the gallbladder. The preoperative diagnosis of HC where no calcifications are present is difficult. Final pathological analysis confirms the presence of hyaline fibrosis [4, 5]. The utility of positron emission tomography with 2- deoxy-2-[f luorine-18] f luoro- D-glucose integrated with computed tomography (18F-FDG PET/CT) in cases of hyalinising cholecystitis has been acknowledged in case reports and have been found to be helpful in establishing the diagnosis. An intense 18F-FDG uptake was noted in cases of HC [6].

It is extremely challenging to diagnose the presence of carcinoma in cases of HC pre-operatively. It is reported that 70% of these cases show no evidence of carcinomatous changes on ultrasound. HC-related carcinomas do not present as mass lesions or as significant wall-thickening. The microscopic analysis is equally challenging for the pathologists. Suspicion should arise if a glandular formation is notes in the hyalinized tissue [1]. Post-operative counselling of patients with HC with associated carcinoma should include their prognosis along

with median survival rate. According to the study by Patel et al., cases with HC-associated carcinoma have statistically worse median survival than patients with the usual gallbladder carcinoma (P = 0.024) [1].

CONCLUSION

This case reports a rare subtype of chronic cholecystitis (hyalinising cholecystitis) diagnosed after a histopathological analysis. Surgeons should be aware of its association with carcinoma gall bladder and hence should alert the pathologist for a thorough microscopic analysis of the removed specimen in cases of suspected intra-operative findings suggestive of HC.

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