CASE REPORT
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Simultaneous Xanthogranulomatous Cholecystitis and Gallbladder Cancer in a Patient with a Large Abdominal Aortic Aneurysm

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There have been reports of the coexistence of abdominal aortic aneurysm (AAA) with intra-abdominal malignancy including gastric, colonic, pancreatic, and renal. We herein report a case of a previously undiagnosed AAA and a presenting complaint consistent with acute cholecystitis. Following cholecystectomy, this was noted to be a rare form of chronic cholecystitis: xanthogranulomatous cholecystitis. There is a known possible association of this uncommon condition with gallbladder cancer. The management of concomitant pathologies can present a real challenge to the multidisciplinary team, especially with large aneurysms.

Keywords: Xanthogranulomatous cholecystitis; Gallbladder neoplasms; Aortic aneurysm, abdominal

INTRODUCTION

Gallstone disease is a commonly encountered condition in surgical practice. Clinical presentation of gallstones varies from silent stones to gallbladder cancer. Rarely, severe cholecystitis may result in extravasation of bile through rupture of Rokitansky-Aschoff sinuses or mucosal ulceration causing severe fibrosis and formation of yellow intramural nodules and xanthogranulomatous reaction known as xanthogranulomatous cholecystitis (XGC) [1-4]. XGC can be easily mistaken for gallbladder cancer both radiologically and macroscopically [1,3,5]. Although there is a known association between gallbladder cancer and XGC, it is not clearly understood.

Computed tomography (CT) has recently become the gold standard investigation of choice in acute abdomen because it is quick, noninvasive, and provides a detailed map of the abdomen. Furthermore, the introduction of multislice CT has increased the diagnostic accuracy of intra-abdominal conditions. However, CT may also detect incidental silent conditions requiring urgent attention such as abdominal aortic aneurysm (AAA).

We herein present a rare case of a patient who presented with signs of acute cholecystitis and gallbladder empyema. Investigations showed evidence of chronic cholecystitis and an incidental large AAA. At surgery, a suspicious small nodule (not seen on CT) was excised. This proved to be a metastasis from gallbladder cancer associated with XGC.

This rare coexistence of pathologies adds a further dimension to the management dilemma of the multidisciplinary team.

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CASE REPORT

A 79-year-old woman presented to the emergency department with a 5-week history of intermittent right upper quadrant and epigastric pain. She did not report any change in her bowel habits or weight loss. Her past medical history included hypertension, myocardial infarction 10 years previously, and bilateral knee replacement.

On examination, she had tachycardia of 100 beats per minute with no fever. Her blood pressure was within normal limits. Abdominal examination revealed a tender mass in the right upper quadrant with a pulsatile epigastric mass. Differential diagnosis by the admitting clinician included a gallbladder mass with a possible AAA. A full blood count and serum biochemistry showed a hemoglobin level of 11.2 g/dL, leukocytosis of 15×10^9/L, C-reactive protein level of 141 mg/L, and normal renal and liver function tests. An urgent CT scan of the abdomen showed evidence of severe cholecystitis with a distended, thick-walled gallbladder, suspected gallbladder empyema, and an incidental 7-cm infrarenal AAA (Fig. 1A). Immediate percutaneous decompression of the gallbladder empyema with a radiologically placed drain revealed 240 mL of pus. Microbiology confirmed positive culture of *Escherichia coli*, for which she received appropriate antibiotics. The patient's condition improved, and she was discharged home. In view of the adjacent AAA, the decision of the multidisciplinary team was to carry out open cholecystectomy followed by staged endovascular aneurysm repair.

At surgery, the gallbladder was adherent to the sur-

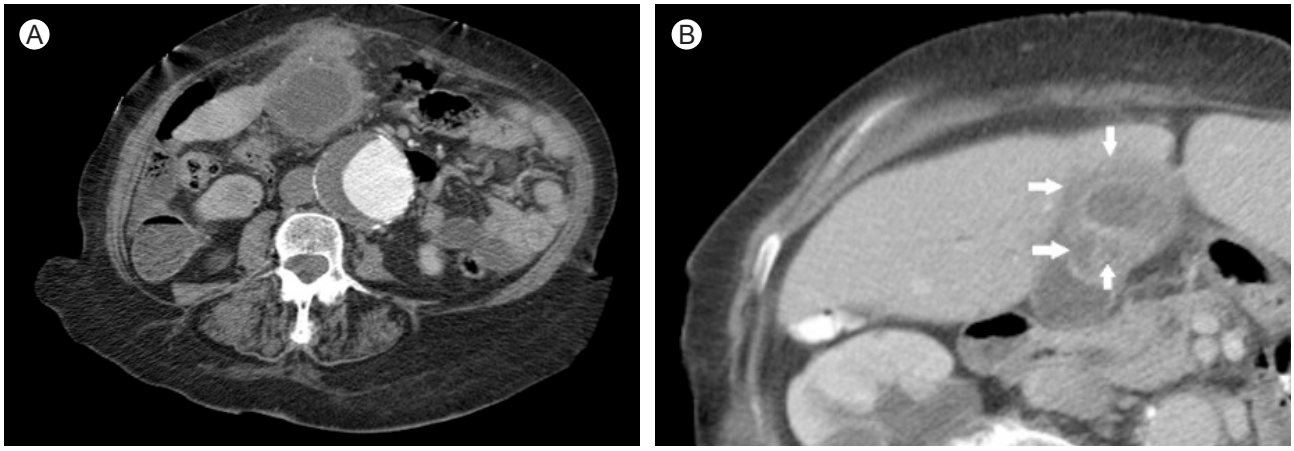

Figure 1. (A) Axial computed tomography (CT) scan of the abdomen showing a large abdominal aortic aneurysm and xanthogranulomatous cholecystitis. (B) CT scan obtained one week after percutaneous drainage of the gallbladder empyema. Note persistence of the hypodense nodules within the gallbladder wall (arrows).

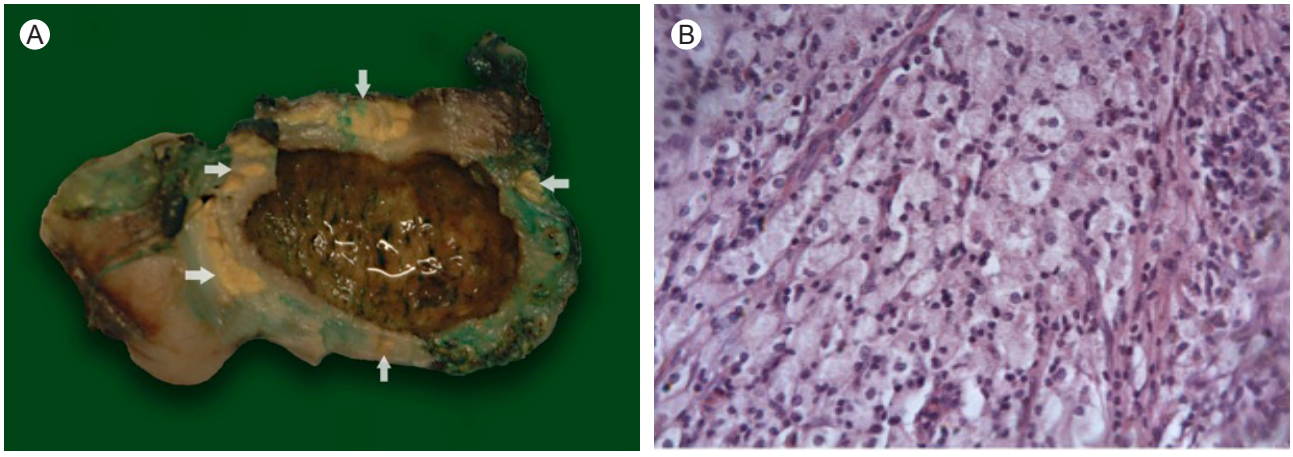

Figure 2. (A) Photograph of the resected gallbladder shows xanthogranulomatous changes within the thick wall gallbladder appearing as yellow nodules (arrows) seen earlier on computed tomography scans. (B) Gallbladder histology shows chronic inflammation with a predominance of foamy macrophages (H&E, × 400).

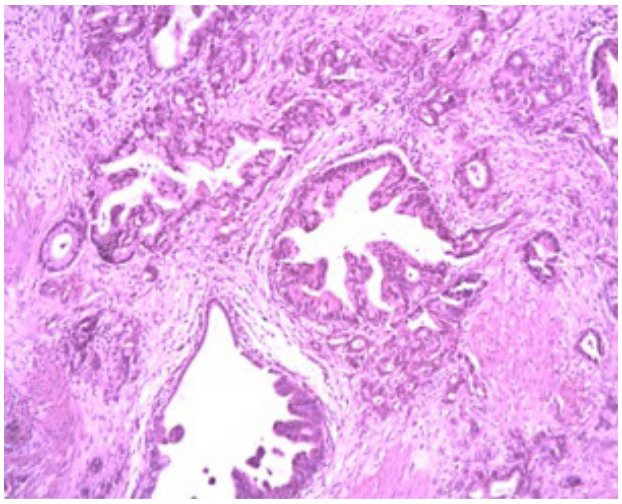

Figure 3. H&E section shows diffusely invasive adenocarcinoma of the gallbladder with dysplastic surface epithelium (H&E, × 200).

rounding tissue with dense liver, duodenal, and bowel adhesions. A small nodule (not clearly seen on CT) was found adjacent to the distended gallbladder. This was also sent to the pathology laboratory. Histology showed XGC (Fig. 2) with adenocarcinoma of the gallbladder. The small nodule proved to be a liver metastasis (Fig. 3). Medical oncologists were then consulted, and she proceeded to chemotherapy.

DISCUSSION

XGC is a rare form of chronic cholecystitis, accounting for 0.7% to 13.2% of resected gallbladder specimens [1,3]. It is thought that chronic inflammation leads to mucosal ulceration and rupture of Rokitansky-Aschoff sinuses with extravasation of lipids from bile, resulting in a xanthogranulomatous reaction within the gallbladder wall characterized by formation of multiple intramural yellow nodules (Fig. 2A) [1-5]. This inflammatory process is often extensive and may extend to adjacent organs, forming dense adhesions with a large mass of inflammatory tissue surrounding the gallbladder [3,5]. The association between XGC and gallbladder cancer has been shown in the literature in small case series and some single case reports. It is estimated that XGC and gallbladder cancer coexist in up to 12% of cases [2].

XGC is often mistaken for carcinoma of the gallbladder both macroscopically during surgery and on CT examinations; thus, radiological differentiation from cancer can be extremely difficult in the presence of severe inflammation [2,6]. Furthermore, the fact that XGC can be associated with gallbladder cancer makes the differentiation even more difficult.

In our patient, the clinical presentation suggested a diagnosis of acute cholecystitis and gallbladder empyema, as confirmed on CT (Fig. 1A) and following percutaneous drainage of gallbladder empyema. Therefore, simple cholecystectomy was carried out as planned; however, advanced gallbladder cancer was discovered during surgery. The single small liver metastatic nodule was very close to the gallbladder. This nodule was less than 1 cm in size and was not shown on CT because of the extensive inflammation surrounding the gallbladder.

Diagnosis of XGC on CT can be difficult; however, the presence of cholelithiasis, thickening of the gallbladder wall of > 3 mm, hypodense nodules within the gallbladder wall, and contrast enhancement of the mucosa with a distinctive presence of a hypodense band around the gallbladder are highly suggestive of XGC [1,4]. Kim et al. [4] suggested that the hypodense nodules may represent abscesses of xanthogranulomas. In our patient, hypodense areas were noted within the gallbladder wall on the initial CT scan. Following percutaneous drainage of the gallbladder empyema, a second CT scan showed that these hypodense areas remained unchanged, confirming that these areas corresponded to the yellow nodules seen macroscopically in the resected specimen (Fig. 1B).

Cholecystectomy, open or laparoscopic, is usually challenging in a xanthogranulomatous gallbladder. Laparoscopic cholecystectomy is often associated with conversion to open surgery in 80% of cases [6,7].

With the increased use of CT as a gold standard imaging modality in abdominal pain, more and more incidental pathologies are identified, some of which prompt urgent attention (e.g., AAA). The increase in the aging population has led to a rise in the prevalence of AAA coexisting with other nonvascular pathologies. It is estimated that 3.5% of patients have an AAA coexisting with intra-abdominal nonvascular disease [8]. The management of concomitant pathology can present a real challenge to the multidisciplinary team, especially with large aneurysms.

It is unclear whether performing simultaneous procedures or a staged approach results in a better outcome.

A study by Fry and Fry [9] concluded that the risk of simultaneous biliary and aortic procedures may subject the patient to a major risk of graft infection and death.

They suggested that a staged operation can be performed safely if the time between the cholecystectomy and subsequent aortic reconstruction is less than 4 months. Another report by Thomas et al. [10] supports the above findings. According to the above studies, the mortality rate due to performance of cholecystectomy in a single operation with AAA repair can reach to 9%. Other reports suggest that combined surgery for both pathologies is safe in selected patients [8]. Even when considering a staged procedure, the order of treatment of both pathologies poses a difficult surgical challenge.

Conflict of interest

No potential conflict of interest relevant to this article is reported.

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